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THE HÆMODYNAMICS OF THE SURGICAL PATIENT UNDER GENERAL ANÆSTHESIA

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THE purpose of this paper is to record and discuss some important circulatory changes which occur in patients subjected to major surgery under general anæsthesia. It is recognized that hæmorrhage (Glasser and Page, 1948), local fluid loss into traumatized tissue (Phemister, 1928; Blalock, 1930), anæsthesia (Zweifach, Hershey, Rovenstine, Lee, and Chambers, 1945), and perhaps noxious nervous stimulation (Crile, 1901; Slome and O'Shaughnessy, 1938) may each, in part, be responsible for the development of these changes, but our main object is to present an overall picture of the circulatory adjustments which take place during the course of major abdominal operations performed under general anæsthesia.

METHODS OF STUDY

Thirty-two adult patients, 18 male and 14 female, undergoing major abdominal operations have been studied. The average age of the patients was 56 years, the range 31 to 80 years.

The patients were sedated, under direction of our anæsthetists, with combinations of morphine, atropine, and scopolamine. The anæsthetics used were combinations of pentothal, curare, cyclopropane, nitrous oxide, and oxygen.

The cardiac index, peripheral blood-flow, blood-pressure, and overall resistance were determined.

Cardiac Index.—This was determined first in the pre-anæsthetic sedated period, and then subsequently during the period of anæsthesia and operation.

A cardiac catheter was introduced into an antecubital vein under local anæsthesia and guided into the right auricle of the heart (McMichael and Sharpey-Schafer, 1944). The catheter was left in position throughout the operation and its patency was maintained by a slow drip of heparinized saline containing 1 ml. of heparin (500 international units) per litre. The maximum amount of saline used never exceeded 600 ml.

Mixed venous blood was withdrawn from the catheter into an oiled syringe, transferred into a bottle containing oil, and stored on ice until analysed for its oxygen content. Arterial blood samples were obtained by direct femoral or brachial artery puncture and were similarly collected and stored. The arterial samples during operation were obtained from either a radial or a brachial artery, but if access proved to be impossible, the arterial oxygen content was deduced from the oxygen capacity of fully saturated venous blood. An arterial saturation of 96 per cent was assumed (Douglas and Edholm, 1949).

The oxygen content of each blood sample was determined on a Van Slyke and Neil manometric apparatus (1924). As shown by Orcutt and Waters (1937), the determinations are not invalidated by the presence of cyclopropane or nitrous oxide in the samples withdrawn during operation; the correction factors remain constant for these two anæsthetic gases in the various solutions used in the technique.

The oxygen consumption rates were measured on a Benedict-Roth spirometer and were determined simultaneously with each blood sampling (Shackman, Graber, and Redwood, 1951).

The cardiac output in litres per minute was computed from the Fick equation,

$$\text{Cardiac Output} = 100 \times \frac{O_2 \text{ consumption c.c. per min.}}{\text{Arterial} - \text{venous } O_2 \text{ content vol. per cent.}}$$

The cardiac index was obtained by dividing the value of the cardiac output by the body surface area, deduced from the height and weight of the patient (Dubois, 1936).

Peripheral Blood-flow.—This was determined at five-minute intervals in 12 of the 32 patients in the pre-anæsthetic sedated period and throughout the period of anæsthesia and operation.

A venous occlusion plethysmograph (Barcroft and Edholm, 1943) was used for the measurements and was applied either to a calf or a foot. Several readings were taken for each determination and the average value used (Lynn and Shackman, 1951).

Blood-pressure.—Blood-pressure recordings were made every 15 minutes by the Korotkow auscultatory method (1905). Mean arterial pressures were computed by halving the sum of the systolic and diastolic readings (Best and Taylor, 1950).

Overall Resistance.—The formula of Apiera (1940) was used to estimate the overall resistance:—

$$R = \frac{P}{F} \times 1332 \times 60,$$

where R = overall resistance in dynes sec. cm.$^{-5}$; F = cardiac output ml./sec.; and P = mean arterial pressure in mm. Hg.

The overall resistance was first determined in each patient from the values obtained during the

pre-anæsthetic sedated period and was accepted as a '*basal*' value. Subsequent increase during operation in the overall resistance thus measured was interpreted as an index of relative vasoconstriction, and subsequent decrease in the overall resistance during operation was interpreted as an index of relative vasodilatation.

RESULTS

Cardiac Index (*Table* 1).—Compared with the values obtained in the pre-anæsthetic sedated period, the cardiac index fell during operation in 78 per cent of patients, rose in 19 per cent, and remained unchanged in 3 per cent.

Table I.—Detailed Results of Oxygen Consumption (c.c./min.), Arterial and Venous Blood-oxygen Content (Vols. Per Cent), Arteriovenous Blood-oxygen Difference (Vols. Per Cent), Cardiac Index (litres/min./m²), and Mean Blood-pressure (mm. Hg) in Pre-anæsthetic Sedated Period and in the Late Phase of Operation.

	Sex	Age	Height (in.)	Weight (lb.)	S/A.	Pre-anæsthetic Sedated Period					During Anæsthesia and Operation					Pre Mean B.P.	Dur. Mean B.P.
						O_2 Cons.	Art. O_2	Ven. O_2	A–V. Diff.	C.I.	O_2 Cons.	Art. O_2	Ven. O_2	A–V. Diff.	C.I.		
1	M.	34	69	132	1·74	208	20·07	18·19	1·88	6·30	215	20·83	17·58	3·25	3·80	106	82
2	M.	50	72	126	1·75	228	14·50	10·99	3·51	3·70	202	16·70	8·67	8·03	1·44	110	122
3	M.	43	58	108	1·40	276	14·73	11·98	2·75	7·10	232	17·90	10·60	7·30	2·30	138	115
4	M.	44	69	136	1·75	325	17·60	15·35	2·25	8·30	197	16·70	13·20	3·50	3·20	100	100
5	M.	54	71	114	1·64	175	17·75	15·50	2·25	4·75	133	19·10	18·00	1·10	7·40	115	112
6	M.	62	68	150	1·57	294	18·40	14·27	4·13	4·50	193	18·30	16·00	2·30	5·30	112	92
7	M.	43	67	138	1·72	310	19·75	15·08	4·67	3·80	181	18·15	16·00	2·15	4·90	140	132
8	M.	73	67	107	1·54	233	13·50	11·20	2·30	6·50	145	15·90	13·64	2·26	4·15	140	90
9	M.	75	66	146	1·75	287	16·90	12·48	4·42	3·70	181	17·83	14·00	3·83	2·70	100	84
10	M.	59	64	139	1·67	203	19·00	14·60	4·40	2·78	162	21·20	18·50	2·70	3·60	95	128
11	M.	47	73	150	1·90	243	17·76	14·90	2·86	4·50	218	18·04	15·10	2·94	3·90	100	120
12	M.	78	69	132	1·72	255	13·70	11·92	1·78	7·70	162	14·93	13·30	1·63	5·50	92	130
13	M.	66	69	108	1·59	217	17·39	15·70	1·69	8·10	196	18·12	15·26	2·86	4·30	105	130
14	M.	45	71	133	1·76	167	16·70	14·75	1·95	4·80	168	18·35	14·25	4·10	2·40	102	110
15	M.	48	68	122	1·65	238	14·12	11·15	2·97	4·80	223	15·58	12·60	2·95	4·60	105	125
16	M.	46	69	164	1·89	267	18·96	17·00	1·96	7·20	267	19·85	18·12	1·73	8·20	165	104
17	M.	38	71	135	1·77	269	21·55	17·51	4·04	3·76	175	19·55	16·40	3·15	3·14	100	98
18	M.	60	73	147	1·87	220	17·38	15·18	2·20	5·35	156	19·35	16·40	2·95	2·80	115	100
	M. Average					246	17·20	14·30	2·90	5·40	189	18·10	14·90	3·20	4·10	113	110
19	F.	68	65	122	1·60	225	13·97	10·20	3·77	3·80	181	14·85	8·09	6·76	1·70	102	102
20	F.	70	61	105	1·43	189	16·99	14·57	2·42	5·50	138	17·15	9·85	7·30	1·40	146	100
21	F.	50	64	116	1·54	257	16·10	14·20	1·90	8·76	138	16·55	14·60	1·95	4·55	120	110
22	F.	50	64	104	1·47	175	17·60	15·60	2·00	5·95	129	17·00	14·75	2·75	3·20	95	118
23	F.	31	60	84	1·28	214	12·95	10·08	2·87	5·80	194	12·95	10·77	2·18	7·00	96	86
24	F.	80	66	103	1·50	148	17·82	12·25	5·57	1·80	155	18·38	13·05	5·33	1·90	115	130
25	F.	72	63	105	1·47	199	14·30	11·55	2·75	4·93	153	16·80	14·20	2·60	4·00	102	105
26	F.	52	62	81	1·30	161	15·58	12·69	2·89	4·30	124	17·08	10·56	6·52	1·46	105	73
27	F.	76	63	99	1·43	230	13·75	10·22	3·53	4·50	138	16·30	13·35	2·95	3·20	130	95
28	F.	74	63	93	1·40	147	16·80	12·65	4·15	2·50	125	16·47	12·05	4·42	2·00	140	94
29	F.	35	67	114	1·60	247	15·20	15·87	2·33	6·60	204	18·70	17·40	1·30	3·40	125	150
30	F.	61	64	109	1·50	195	18·40	12·77	5·63	2·30	169	17·82	9·87	7·95	1·40	120	115
31	F.	57	60	136	1·56	271	15·40	13·00	2·40	7·20	217	15·45	10·23	5·22	2·67	120	100
32	F.	56	65	134	1·65	194	17·50	15·00	2·50	4·70	140	18·30	14·90	3·40	2·50	123	100
	F. Average					204	16·10	12·90	3·20	4·90	157	16·70	12·40	4·30	2·90	117	106

The average cardiac index of the 32 patients in the pre-anæsthetic sedated period was 5·4 litres/min./m² (range 2·8 to 8·3) in 18 males, and 4·9 (range 1·8 to 8·8) in 14 females. The average cardiac index of the 32 patients during operation was 4·1 (range 1·4 to 8·2) in the males and 2·9 (range 1·4 to 7·0) in the females.

The average cardiac index during operation of those 25 patients whose cardiac index fell was 3·4 (range 1·4 to 5·5) in 13 males and 2·6 (range 1·4 to 4·6) in 12 females; the average fall in cardiac index in the males was equivalent to 37 per cent of the average pre-anæsthetic value, and the average fall in cardiac index in the females was equivalent to 47 per cent of the average pre-anæsthetic value.

The average cardiac index during operation of the 6 patients whose cardiac index rose was 5·9 (range 3·6 to 8·2) in 5 males, and 7·0 in a single female; the average rise in cardiac index in the males was equivalent to 7 per cent of the average pre-anæsthetic value, and the average rise in cardiac index in the single female was equivalent to 43 per cent of the respective pre-anæsthetic value.

No change in cardiac index was noted in a single female patient.

Table II.—Calf and Foot Blood-flows (ml./100 c.c./min.), Cardiac Indices (litres/min./m²) and Mean Arterial Pressures (mm. Hg) in Pre-anæsthetic Sedated Phase, Period of Peak Limb Flow, and Late Phase of Operation

No.	Sex	Age	Resting Flow	B.P.	C.I.	Peak Flow	B.P.	C.I.	Final Flow	B.P.	C.I.	Part Studied
2	M.	50	1·0	110	3·7	3·0	105	2·4	1·0	122	1·3	Calf
25	F.	72	6·0	102	5·0	15·0	97	3·8	2·8	105	4·2	Calf
6	M.	62	2·5	112	4·5	15·0	115	—	2·0	92	5·3	Calf
30	F.	64	1·5	120	2·3	5·9	110	2·1	1·0	115	1·4	Calf
29	F.	35	1·4	125	6·6	5·3	125	—	2·0	150	3·4	Calf
4	M.	44	3·5	100	8·3	8·0	110	—	2·0	100	3·2	Calf
3	M.	43	1·7	138	7·1	5·0	135	—	2·0	115	2·3	Calf
			2·5			8·2			2·3			Average calf flow
19	F.	68	1·3	102	3·8	2·5	75	2·3	0·7	102	1·7	Foot
20	F.	70	2·2	146	5·5	8·5	150	4·0	2·0	100	1·4	Foot
24	F.	80	2·0	115	1·8	5·0	112	2·2	1·0	130	1·9	Foot
16	M.	46	2·8	165	7·2	13·3	125	—	4·9	104	8·2	Foot
17	M.	38	2·0	100	3·8	12·5	100	—	1·5	98	3·1	Foot
			2·1			8·4			2·0			Average foot flow

Peripheral Blood-flow (*Table II*).—This was determined in 12 patients. In the pre-anæsthetic sedated period the average calf blood-flow was 2·5 ml./min. per 100 c.c. of tissue, and the average foot flow 2·1.

The calf and the foot flows were found to increase in all 12 patients as soon as anæsthesia was induced (*Fig.* 239). The flows became maximal after about 30 minutes had elapsed and then the calf flows

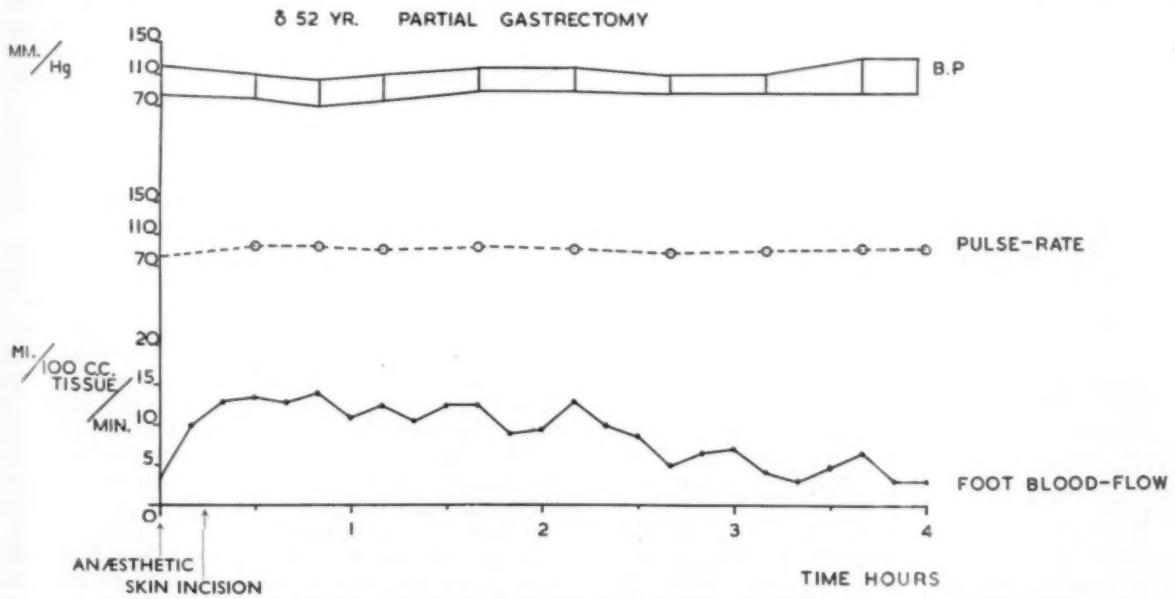

Fig. 239.—Increased limb flow beginning at induction of anæsthesia and becoming maximal within forty minutes. Blood-pressure and pulse-rate remain virtually unchanged.

averaged 8·2 ml. and the foot 8·4 ml.; these values were 230 per cent and 300 per cent respectively greater than the corresponding values obtained in the pre-anæsthetic sedated period.

During the late period of the operations the calf and foot flows both returned approximately to their corresponding pre-anæsthetic values in all 12 patients.

The cardiac index was measured in 6 of the 12 patients during the period of increased limb flow; in 5 the cardiac output was found at this time to be less than the value obtained in the corresponding pre-anæsthetic phase, and in the remaining single case it was found to be somewhat greater. The average decrease of cardiac index in the 5 patients in which it fell during the period of increased limb flow was equivalent to 28 per cent of the average pre-anæsthetic cardiac index, while the single increase was equivalent to 22 per cent of the corresponding pre-anæsthetic cardiac index.

Table III.—Relation between Cardiac Index and Overall Resistance in Pre-anæsthetic Sedated Phase and Late Period of Operation

	Fall of Cardiac Index	Rise of Cardiac Index	No Change of Cardiac Index	
Overall vasoconstriction	22	0	1	72 per cent
Overall vasodilatation	1	5	0	19 per cent
No change of overall resistance	2	1	0	9 per cent
	78 per cent	19 per cent	3 per cent	

The cardiac index was determined in all 12 patients during the late period of operation when the limb flows had fallen from their peak values and had returned approximately to their pre-anæsthetic values. In 9 the cardiac index was then found to be less than the corresponding pre-anæsthetic value, in 2 it was more, and in 1 it was virtually unchanged; the average decrease was equivalent to 47 per cent of the average pre-anæsthetic cardiac index, while the average increase was equivalent to 13 per cent of the average pre-anæsthetic cardiac index.

Blood-pressure (*Table I*).—In the pre-anæsthetic sedated period, at the time of the primary determinations of cardiac index, the average mean blood-pressure was 113 mm. Hg (range 92 to 165) in 18 male patients, and 117 (range 95 to 146) in 14 females.

At the time of the subsequent final determinations of cardiac index during operation, the average mean blood-pressure was 110 mm. Hg (range 82 to 130) in the males, and 106 (range 73 to 150) in the females.

The mean blood-pressure had fallen in 10 males and in 9 females, had risen in 7 males and 4 females, and had remained unchanged in a single male and a single female.

Overall Resistance.—When computed from the formula of Apiera (1940), the average value for overall resistance in the 32 patients in the pre-anæsthetic sedated phase was found to be 1300 dynes sec. cm.$^{-5}$ This value resembles that recorded by Cournand, Riley, Bradley, Breed, Noble, Lausen, Gregersen, and Richards (1943) for non-anæsthetized subjects; in general, the overall resistance in our male patients in the pre-anæsthetic sedated phase was somewhat lower than the average value recorded by Cournand and others (1943), while in our female patients it was somewhat higher.

The average value for overall resistance in the 32 patients in the late period of operation was calculated to be 1910, the males averaging 1510, and the females 2420.

Twenty-three patients (72 per cent) had reacted to anæsthesia and operation by an increase of overall resistance, 6 (19 per cent) had reacted by a decrease of overall resistance, and 3 (9 per cent) showed no change. Correlation between changes of overall resistance and changes of cardiac index are seen in *Table III*. Of the 23 patients who developed an increase of overall resistance during operation, 22 were found to have a concomitant fall in cardiac index, and in a single patient the cardiac index was unchanged. Of the 6 patients who developed a decrease of overall resistance during operation, 5 were found to have a concomitant rise in cardiac index and 1 a fall. Of the 3 patients in whom the overall resistance was unchanged, 2 were found to have a fall in cardiac index and one a rise.

DISCUSSION

The average value of our primary determinations of cardiac index in patients in the pre-anæsthetic sedated phase is somewhat greater than the values recorded by Cournand, Riley, Breed, Baldwin, and Richards (1945), McMichael and Sharpey-Schafer (1944) and Stead, Warren, Merrill, and Brannon (1945) for 'normal' hospital personnel (*Table IV*). This possibly reflects the degree of apprehension experienced by our patients, but is also partly the result of our inclusion of some exceptionally high values (*Cases* 3, 4, 12, 13, 16, 21, 31). The corresponding average values for blood-pressure and limb flow, however, show no significant variation from 'normal'.

The studies have shown that circulatory re-adjustments occur during general anæsthesia and major abdominal surgery and that these re-adjustments begin as soon as anæsthesia is induced. The maximal increases of limb flow, which develop within about 30 minutes after induction, average almost 270 per cent more than the average pre-anæsthetic basal limb flows and occur in the skin and muscle of all four limbs. Since these changes are representative of those occurring in the whole of the body skin and musculature, and since about 960 ml./min. of blood normally circulates through these tissues (Glasser, 1950), it may be concluded that the total increase of blood-flow in the skin and muscle of the body during the early period of anæsthesia is in the region of 2500 ml./min.

Since the increases in skin and muscle flow occur despite a virtually unchanged blood-pressure and in the presence of a decreased cardiac output (5 of 6 patients whose cardiac output was measured in this period), it may be concluded that compensatory vasoconstriction has developed elsewhere in the body. We have no direct evidence in humans as to the site of this compensatory phenomenon, but it

may take place in the pulmonary, cerebral, renal, and splanchnic circulations.

Pulmonary vasoconstriction, without a concomitant rise of pressure in the pulmonary artery, during general anæsthesia would result in a decreased return of blood to the left auricle, a fall in cardiac output, and a fall in systemic blood-pressure. To maintain an unaltered systemic blood-pressure in the presence of a reduced cardiac output there must be an associated vasoconstriction in the systemic circulation. The fact that the systemic blood-pressure remained virtually unchanged in our patients in the early period of anæsthesia and operation therefore suggests that pulmonary vasoconstriction alone plays little or no part in the production of the compensatory changes that we have postulated.

The cerebral circulation in man is almost certainly modified during general anæsthesia. Kety (1949) has stated that there is a correlation between cerebral blood-flow and cerebral oxygen consumption in monkeys, and Schmidt (1945) has shown that cerebral metabolism is decreased under pentothal anæsthesia. Since, however, the average cerebral blood-flow in the adult human is only about 760 ml./min. (Glasser, 1950), it is clear that any variation from normal could not be great enough alone to counterbalance the great increase in limb flow that we have recorded: even a complete shut-down of the cerebral circulation would itself be inadequate for maintenance of the systemic blood-pressure in such circumstances.

Comparatively little is known about the effects of general anæsthesia and surgery on the renal circulation. Coller, Rees, Campbell, and Iob (1943) studied 5 patients by the Homer Smith (Smith, Goldring, and Chasis, 1938) techniques of renal clearance, and we ourselves have made one such study. The available evidence suggests that during the period when the limb flows are increased, the effective renal blood-flow is also usually somewhat increased and that the variations from normal which may occur are not large enough alone to effect any significant change in the hæmodynamics of a patient in the early period of anæsthesia and operation.

The splanchnic bed is the most likely site of the compensatory vasoconstriction. It has been shown in animals (Müller, 1905; Müller and Siebeck, 1907; Müller and Veiel, 1909) that there is reciprocity between the peripheral blood-flow and the intra-abdominal blood-flow, and the observation has been recently confirmed in humans (Grayson, 1949) by the use of thermocouples to measure simultaneously temperature changes of the limbs and exteriorized bowel. Splanchnic vasoconstriction has been directly observed in dogs in the vessels of exteriorized omentum after induction of anæsthesia by cyclopropane or nitrous oxide (Zweifach and others, 1945), and it has also been shown (Selkurt, Alexander, and Patterson, 1947), that splanchnic vasoconstriction develops in dogs subjected to bleeding under general anæsthesia. There is therefore good experimental evidence to support a contention that splanchnic vasoconstriction may occur in humans during operation under general anæsthesia.

The circulatory adaptation which begins in patients immediately after induction of anæsthesia has been found to be remarkably constant. Vasodilatation in the limb vessels develops along with reciprocal vasoconstriction, probably in the splanchnic bed, with the blood-pressure remaining virtually unaltered and the cardiac output usually decreased. The fact that these changes occur so rapidly after induction of anæsthesia and before any really active surgery has begun leads to the assumption that they result primarily from the administration of the anæsthetic.

The modified circulatory pattern thus described persists until some further stimulus is produced by prolonged anæsthesia and operation. The limb flows then decrease and there is vasoconstriction in the limb vessels when compared with their state in the period of the peak limb flows. This subsequent change in the state of the limb vessels is associated

Table IV.—Present Authors' Values compared with those obtained by Cournand and others (1945), McMichael and others (1944), and Stead and others (1945)

	Cournand and others	McMichael and others	Stead and others	Present Authors	
	Hospital 'Normal' patients	'Normal' subjects	Excited subjects	Pre-anæsthetic Sedated Phase	Late Operation
Oxygen consumption	270	240	157 M^2	246 M. 204 F.	189 M. 157 F.
Arterial oxygen	14·8 M. and F. (96 per cent saturation of 15·4)		18·0	17·2 M. 16·1. F	18·1 M. 16·7 F.
Venous oxygen	10·5 M. 10·9 F.			14·3 M. 12·9 F.	14·9 M. 12·4 F.
A–V. difference	4·3 M. 3·9 F.	4·5	3·1	2·9 M. 3·2 F.	3·2 M. 4·3 F.
Capacity	15·4 Hb 11·5g.				
Cardiac index	3·72 M. 2·99 F.	3·1 (Assume Average) (S.A. = 1·69)	5·5	5·4 M. 4·9 F.	4·1 M. 2·9 F.

with two alternate and distinct overall circulatory patterns. The majority of our patients developed a fall in cardiac output and an overall increase in vasomotor tone, while a small minority developed a rise in cardiac output and an overall decrease in vasomotor tone (*Figs.* 240, 241).

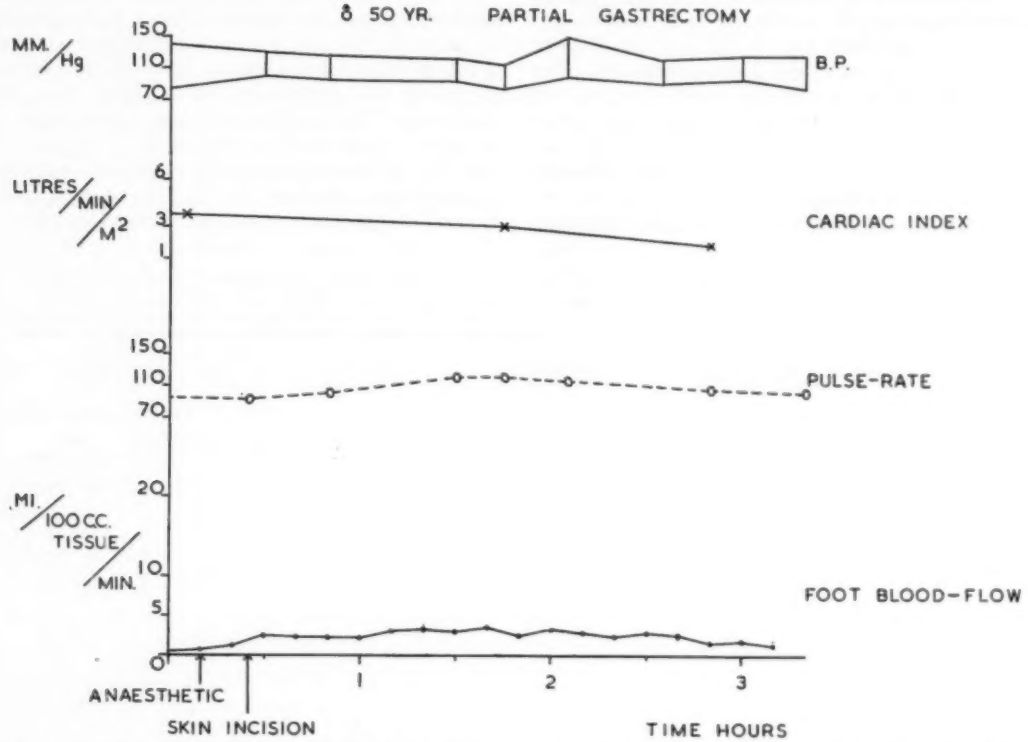

FIG. 240.—Fall of cardiac index during operation. Blood-pressure and pulse-rate remain virtually unchanged. Limb flow falls from peak level towards level of pre-anæsthetic sedated phase.

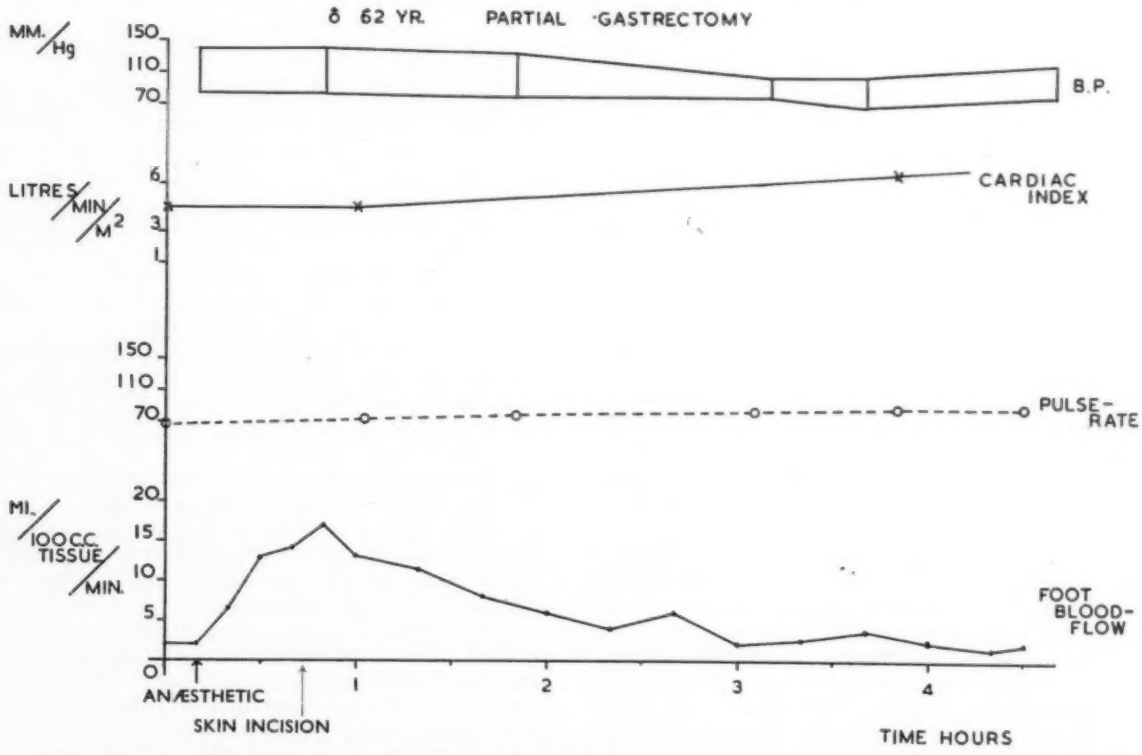

FIG. 241.—Rise of cardiac index during operation with slight fall of blood-pressure and unchanged pulse-rate. Limb flow returns to level of pre-anæsthetic sedated phase.

Since the degree of relative vasoconstriction in the limb vessels which develops in the late stages of operation is not paralleled by changes in the systemic blood-pressure and cardiac output, it is concluded that a degree of splanchnic vasodilatation, relative to the state of the splanchnic vessels during the peak limb flow, has occurred simultaneously. The splanchnic vessels, however, are still relatively constricted when compared with their state in the pre-anæsthetic sedated period. The limb vessels, on the other hand, although constricted relative to the state of the limb vessels in the period of the peak limb flows, are slightly dilated when compared with their state in the pre-anæsthetic sedated period. (*See Note* 1.)

In the small group of patients whose cardiac output rose and overall resistance fell, the splanchnic vessels are dilated in the late period of operation relative to both the period of peak limb flows and the pre-anæsthetic sedated phase. The limb vessels, on the other hand, like those of the previous group, although relatively constricted when compared with their state in the period of peak limb flows, are slightly dilated when compared with their state in the pre-anæsthetic sedated phase. (*See Note* 2.)

Note 1.—We have used the Apiera (1940) formula $R = \frac{P}{F} \times 1332 \times 60$ to calculate resistances in dynes sec. cm.$^{-5}$.

When L = flow in total body muscle and skin (ml./min), CO = cardiac output (ml./min), R_1 = resistance in total body muscle and skin, R_{r+s} = resistance in the rest of the body, and P = mean arterial pressure (mm. Hg),

then $$R_1 = \frac{P}{L} \times 1332 \times 60$$

and $$R_{r+s} = \frac{P}{CO - L} \times 1332 \times 60.$$

If an average calf flow of 2·5 ml./100 g. is accepted as representing muscle flow (*Table II*), and there is 24,000 g. of body muscle (Glasser, 1950), the total body muscle flow is

$$24{,}000 \times \frac{2{\cdot}5}{100} = 600 \text{ ml./min. (per } 1{\cdot}7 \text{ m}^2).$$

And since about 200 ml. of blood normally circulate through the body skin (Glasser, 1950), the average total resting blood-flow through all body muscle and skin may be considered to be in the region of 800 ml./min.

Considering *Cases* 2, 19, 20 24, 25, 30, in whom the cardiac output determinations were available for all stages, and in whom the surface area averaged 1·54, we get,

(i) *In the pre-anæsthetic sedated phase:*—

$$R_1 = \frac{116}{800 \times \frac{1{\cdot}54}{1{\cdot}7}} \times 1332 \times 60 = 12{,}900 \text{ (dynes sec. cm.}^{-5})$$

and

$$R_{r+s} = \frac{116}{5700 - 720} \times 1332 \times 60 = 1930 \text{ (dynes sec. cm.}^{-5}).$$

Since the average increases in the muscle and skin flow were respectively 230 and 300 per cent (*Table II*) we get,

(ii) *In the early period of anæsthesia when the limb flows are maximal:*—

$$R_1 = \frac{108}{2440} \times 1332 \times 60 = 3540 \text{ (dynes sec. cm.}^{-5})$$

and

$$R_{r+s} = \frac{108}{4300 - 2440} \times 1332 \times 60 = 4700 \text{ (dynes sec. cm.}^{-5}).$$

Since the average flow in muscle and skin returns approximately to the values present in the pre-anæsthetic sedated phase (*Table II*) we get,

(iii) *In the late period of anæsthesia and operation:*—

$$R_1 = \frac{112}{720} \times 1332 \times 60 = 12{,}400 \text{ (dynes sec. cm.}^{-5})$$

and

$$R_{r+s} = \frac{112}{3170 - 720} \times 1332 \times 60 = 3660 \text{ (dynes sec. cm.}^{-5}).$$

Note 2.—Using the values obtained in *Case* 6 as an example we get,

(i) *In the pre-anæsthetic sedated phase:*—

$$R_1 = \frac{112}{750} \times 1332 \times 60 = 12{,}000 \text{ (dynes sec. cm.}^{-5})$$

and

$$R_{r+s} = \frac{112}{7100 - 750} \times 1332 \times 60 = 1410 \text{ (dynes sec. cm.}^{-5}).$$

(ii) *In the early period of anæsthesia when the limb flows are maximal* (assuming the cardiac output to be the same as in (iii) :—

$$R_1 = \frac{115}{3900} \times 1332 \times 60 = 2320 \text{ (dynes sec. cm.}^{-5})$$

and

$$R_{r+s} = \frac{115}{4400} \times 1332 \times 60 = 2010 \text{ (dynes sec. cm.}^{-5}).$$

(iii) *In the late period of anæsthesia and operation:*—

$$R_1 = \frac{92}{645} \times 1332 \times 60 = 11{,}400 \text{ (dynes sec. cm.}^{-5})$$

and

$$R_{r+s} = \frac{92}{7655} \times 1332 \times 60 = 970 \text{ (dynes. sec. cm.}^{-5}).$$

The reason for these divergent circulatory reactions is not clear, but somewhat similar findings have been reported (Remington, Hamilton, Cadell, Boyd, and Hamilton, 1950) to occur in dogs subjected to repeated small bleedings under morphine and pentothal anæsthesia. Remington and others (1950) postulated that the opposite responses reflect the relative levels of resistance and flow in the control periods. When resistance is low and flow is high, that is when there is a relatively low degree of vasomotor tone, subsequent bleeding is followed by a decline in flow. Conversely, when resistance is high and flow is low, that is when there is a relatively high degree of vasomotor tone, bleeding is followed by an increased flow.

Applying this postulate to our patients, it seemed at first that those with a relatively low index of overall resistance in the pre-anæsthetic phase subsequently reacted to anæsthesia and operation by a fall in cardiac output, whereas those with a relatively high degree of resistance in the pre-anæsthetic phase subsequently developed an increased cardiac output. Independent statistical analysis, however, throws doubt on the significance of these particular findings.

The significance of the circulatory changes which we have described may be profound. Splanchnic vasoconstriction decreases the liver blood-flow (Wiggers, Opdyke, and Johnson, 1946 ; Selkurt and others, 1947), and there is reason to believe that a direct relation exists between impairment of liver

blood-flow and function, and the development of irreversible shock (Frank, Seligman, and Fine, 1946; Shorr, Zweifach, Finchgott, and Baez, 1951; Delorme, 1951). There is some evidence that varying degrees of liver dysfunction may occur in humans undergoing surgery under general anæsthesia (Boyce and McFetridge, 1938; Tagnon, Robbins, and Nichols, 1948), and it is possible that splanchnic vasoconstriction is partly responsible (*Fig.* 242).

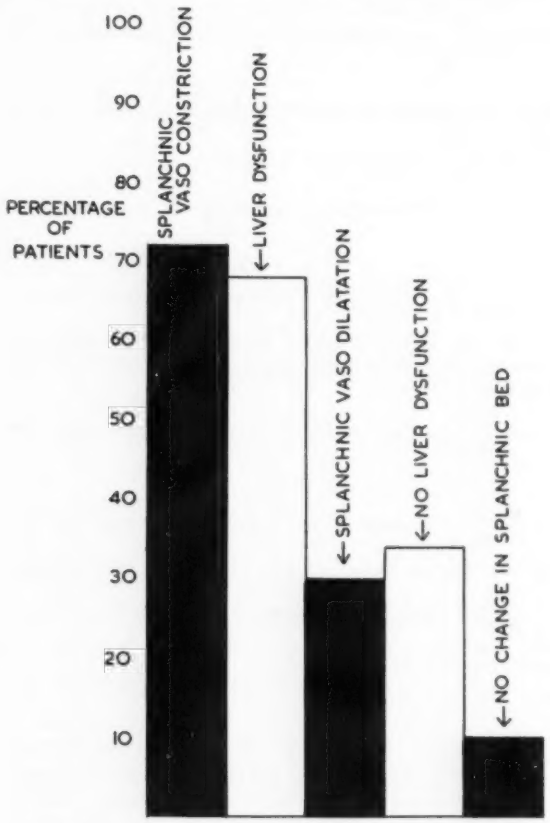

FIG. 242.—Similarity between incidence of splanchnic vasoconstriction and incidence of liver dysfunction; 20 patients studied by Tagnon and others (1948); 32 patients studied by present authors.

It is perhaps premature to deduce any practical applications from the results of our studies, but it is clear that excessive blood-loss will accentuate the hæmodynamic changes and prolonged operating time will increase their duration.

SUMMARY

1. Some aspects of the circulation have been studied in 32 adult patients undergoing major abdominal surgery under general anæsthesia.

2. Cardiac outputs were determined by cardiac catheterization, skin and muscle blood-flows by water plethysmography, and blood-pressures by cuff sphygmomanometry.

3. Measurements were made first in the pre-anæsthetic sedated phase and then subsequently during operation.

4. Changes in vascular resistance in skin and muscle, and in the rest of the body, were computed from the values so obtained.

5. It has been found that major circulatory adjustments begin as soon as general anæsthesia is induced and that practically half of the cardiac output very soon becomes distributed to the body skin and musculature.

6. This redistribution of blood has been found to occur usually in the presence of a slight fall in cardiac output and a virtually unchanged blood-pressure, and is explained on the basis of a profound vasodilatation of the blood-vessels in the skin and muscle.

7. It has been deduced that compensatory vasoconstriction, most probably in the splanchnic bed, accompanies these early circulatory changes.

8. Prolongation of anæsthesia and operation was found to be accompanied by yet further circulatory changes. In the late stage of operation, the skin and muscle blood-flows decreased and returned approximately to the levels present in the pre-anæsthetic sedated phase, while the cardiac output fell appreciably in the majority of patients and rose in a small minority.

9. Calculation of resistances in this late stage showed that overall vasoconstriction had developed in 72 per cent of patients and overall vasodilatation in 19 per cent. (No change in overall resistance was found in the remaining 9 per cent of patients.)

10. It has been deduced that splanchnic vasoconstriction was still present in the late phase of operation in the major group of patients.

11. The effects of prolonged splanchnic vasoconstriction are discussed and attention is drawn to the implications of a reduced splanchnic and liver blood-flow in patients undergoing major abdominal surgery.

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THE DIAGNOSIS OF INTESTINAL OBSTRUCTION IN THE NEWBORN

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UNTIL recently intestinal obstruction in the newborn resulted in a high mortality. Latterly, however, there have been indications that many affected babies can be saved if the diagnosis is made early enough and if the modern developments in the management of these cases are applied.

The following is an account of eighteen cases of intestinal obstruction presenting within a month of birth. They occurred over a period of three years in a children's hospital and most of them were personally observed. Methods of diagnosis are discussed. It is important to attempt to determine the cause of the obstruction before operation. Failure to recognize the nature and characteristic features of the various types of ileus may lead to grave errors in procedure, or to injurious prolongation of the operation.

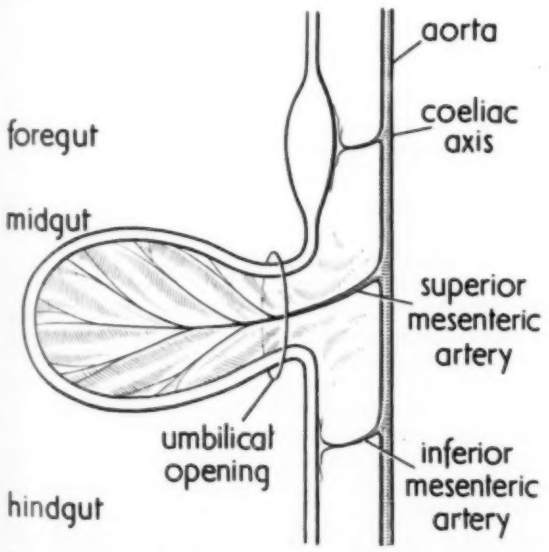

FIG. 243.—Lateral view of gut at about ninth week of fœtal life, showing midgut protruding into base of umbilical cord.

Most of the obstructing mechanisms are peculiar to this period and are developmental in origin.

Anomalies of Intestinal Rotation.— Dott (1923) gave a masterly account of the embryology of intestinal rotation and described the anomalies which occur. Ladd and Gross (1941) have clarified the mechanisms which cause intestinal obstruction in early life. Between the sixth and tenth weeks of fœtal life the intestinal tract grows more rapidly than does the abdominal cavity, and the midgut prolapses into the base of the umbilical cord (*Fig.* 243). The abdominal portion of the foregut forms the stomach, and the duodenum as far as the biliary papilla. At an early date bulging of the greater curvature of the stomach occurs ; this results

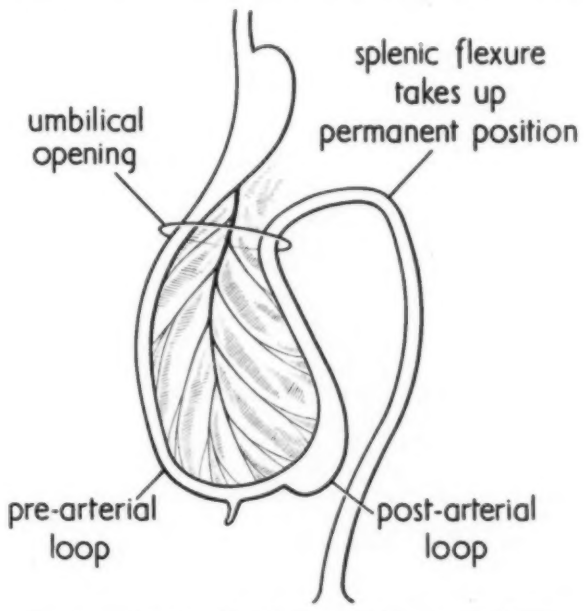

FIG. 244.—Anterior view of gut at tenth week. Duodenum has moved to right, initiating the process of rotation.

in the obliquity of its long axis. The duodenum is pushed to the right (*Fig.* 244). This movement begins the counter-clockwise rotation of the midgut which now occurs. The hind end of the midgut, which corresponds to the distal end of the adult

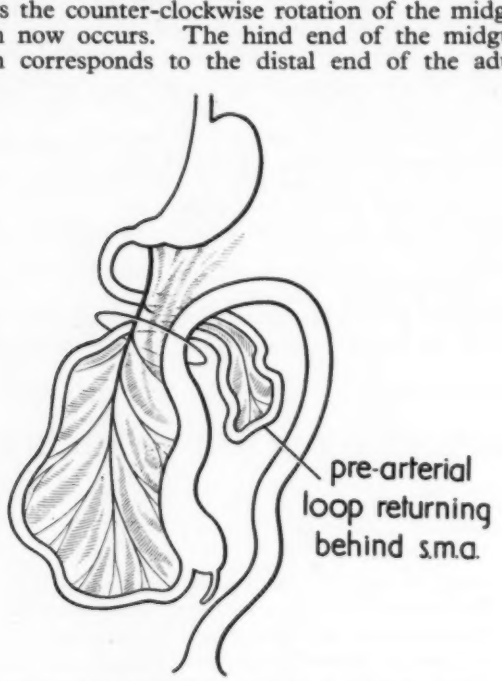

FIG. 245.—At the end of the tenth week, the pre-arterial loop of the midgut is returning to the abdomen. It passes to the left behind the arterial axis.

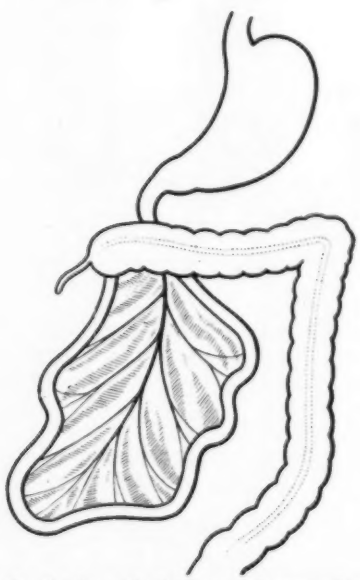

FIG. 246.—At the end of the second stage of rotation the whole gut has returned to the abdominal cavity, but the cæcum is still suspended by a mesentery in the subpyloric region. Rotation often stops with the bowel in this position.

transverse colon, is pushed over to the left where it takes over its permanent position in the left upper quadrant. It also becomes relatively fixed and lies close to the duodenum. Rotation takes place around this duodeno-colic isthmus, and then at the end of the tenth week the midgut begins to return to the duodenum. The proximal part (small intestine) returns first and has rotated so far that it travels behind the superior mesenteric artery and over to the left (*Fig.* 245). Lastly the cæcum is reduced and comes to lie anterior to the small intestine and mesenteric artery. Now the colon is lying across the duodenum and artery (*Fig.* 246).

In the third stage of rotation the cæcum descends into the right flank and applies itself to the posterior abdominal wall. The mesenteries of the ascending colon and cæcum fuse with the posterior parietal peritoneum.

The commonest anomaly to produce symptoms at birth is that due to interference with the end of the second and with the third stage of rotation. The cæcum lies in the subpyloric region and is suspended by a mesentery. As a result the ileo-cæcal junction is approximated to the duodeno-jejunal flexure and the cæcum is quite mobile. The

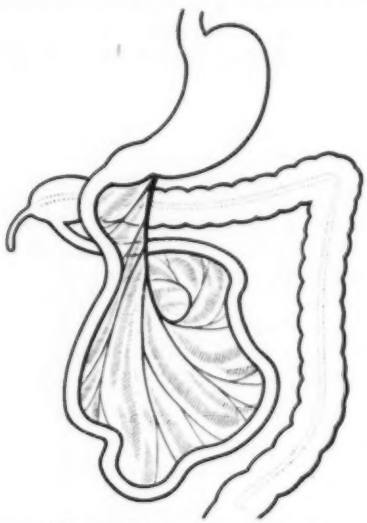

FIG. 247.—'Reversed' rotation. The post-arterial loop has returned to the abdomen first and has passed to the right behind the arterial axis.

ideal conditions for a volvulus are produced, and the small bowel rotates in a clockwise manner with the superior mesenteric artery as an axis.

Alternatively, the sequence of the return of the bowel may be reversed. (*Fig.* 247.) If the umbilical opening is large the cæcum and ascending colon return to the abdomen first. They are deflected to the left behind the superior mesenteric artery and the small bowel follows. Eventually, the duodenum comes to lie anteriorly to the arterial axis.

Both these conditions, deficient rotation and 'reversed' rotation, may be associated with volvulus.

Ladd and Gross (1941) pointed out that a second obstructing lesion is often present. Peritoneal bands are found running from the cæcum to the right side of the abdomen, across the second part of the duodenum. They produce sufficient intrinsic pressure to cause dilatation of the proximal duodenum which in turn results in ptosis and kinking of the duodenum over the band, aggravating the obstruction. Alternatively the duodenum may be pressed upon by the overlying cæcum.

At operation the collapsed and cyanosed loops of small bowel present. They ought to be delivered from the wound and then untwisted in a counter-clockwise direction, generally through 360°. When

the volvulus is corrected the duodenal region must be examined, and if obstructing bands are present they are divided. The cæcum is then free to be pushed over to the left side of the abdomen. It appears to be preferable to have the colon on the left side rather than to attempt to establish the normal anatomical arrangement by fixing the cæcum in the right lower quadrant.

Other lesions may sometimes be found in addition. Madding, Everhart, and Heath (1950) and others have described cases of intestinal malrotation associated with duodenal atresia, and *Case* 6 is an example of this anomaly. Hence it appears advisable to exclude the possibility of duodenal atresia too, by guiding the inlying gastric catheter through the duodenum into the jejunum.

Intestinal Atresias.—Various theories have been suggested to explain the occurrence of intestinal atresias. The theory of persistence of physiological epithelial occlusion is the one most commonly accepted. The epithelium of the gut proliferates considerably in the second fœtal month and the lumen becomes blocked. At a later date the bowel recanalizes. It is suggested that atresias represent a failure of this latter process.

This mechanism does not explain the type where there is a complete gap in the intestine accompanied by a V-shaped defect in the mesentery. In other varieties there may be a gap joined by a solid cord, an annular constriction, or a diaphragm across the lumen.

The duodenum is a fairly common site for intestinal atresias. Most of them are below the ampulla of Vater so that the vomit is bile-stained and the meconium light in colour. Occasionally the common bile-duct is hypertrophied and dilated, as well as the duodenum, and the baby is jaundiced. There is little distension of the abdomen when the obstruction is high.

Atresias of the small intestine are commonest in the ileum. They are accompanied by considerable distension from birth. Colonic atresias, apart from malformations of the rectum and anus, are rare. Atresias are often multiple, and after making an anastomosis it is essential to examine the bowel below. Also it must be remembered that cases of imperforate anus may have another atresia at a higher level.

Neuromuscular Dysfunction. — Zuelzer and Wilson (1948) showed that neuromuscular dysfunction, presumably due to absence of ganglion cells from the wall of the bowel, can produce obstructions at any level in the colon and occasionally in the ileum. They reported two cases of the latter in their series of eleven cases, and a further case was described by Bodian, Carter, and Ward (1951), and two cases are included in this series. The absence of ganglion cells generally affects the rectum and lower sigmoid, giving rise to the condition described by Hirschsprung in 1886. Only a small proportion of these cases present with symptoms in the newborn period severe enough to justify surgical treatment immediately. Various authors have demonstrated the strong familial tendency in this disease, which may be of value in differential diagnosis.

Meconium Ileus. — In this condition the meconium is so inspissated and tenacious that blockage of the terminal ileum results. The distal ileum is filled with hard, inspissated, grey fæces and proximally it is grossly distended and hypertrophied. The contents of the tapering segment of the bowel between the constricted and dilated areas are dark-coloured and peculiarly viscous and sticky. The colon is small, contracted and worm-like. It appears that this condition was commonly called microcolon and was regarded as the actual cause of the intestinal obstruction. Whenever this lack of development of the colon occurs, a search must be made for an obstructing agent in the terminal ileum.

Meconium ileus is generally associated with fibrocystic disease of the pancreas, and the abnormal state of the meconium is believed to be due to the absence of trypsin from the bowel. Rapoport and Buchanan (1950) have shown that meconium is the residue of the mucous secretions of the active alimentary tract. In meconium ileus it contains mucoproteins, whereas normal meconium contains mucopolysaccharides and nitrogen compounds soluble in water, which result from the digestion of the mucoproteins by trypsin. Mucoproteins are more viscous than mucopolysaccharides.

Miscellaneous.—Besides the four main groups of obstructing lesions various others occur less commonly. External and internal hernia may strangulate, or a volvulus may occur in association with a Meckel's diverticulum. An annular pancreas may be found, reduplications of the bowel occasionally cause symptoms soon after birth. In this series an adhesion, apparently due to an inflammatory lesion of the small intestine, was found.

DIAGNOSIS

It is clear that the majority of these obstructing lesions are remediable, and that the poor results of surgery hitherto have been due to delay in diagnosis, inadequate pre-operative preparation, and sometimes to a failure to realize the real nature of the obstruction at operation.

The diagnosis should present no difficulty. The cardinal symptoms are bilious vomiting, abdominal distension, and abnormalities of bowel action. Once the suspicion of intestinal obstruction is raised it is easy to confirm or disprove the suspected diagnosis by plain radiography. Wangensteen and Lynch (1930) showed experimentally that X-ray evidence was present within four or five hours of producing experimental obstruction in dogs, and Koch (1944) showed that a definite diagnosis could be made in 111 out of 119 cases of obstruction by X-ray examination.

Some degree of vomiting during the first thirty-six hours of life occurs in about 90 per cent of babies. Swallowed amniotic fluid, vaginal secretion, and blood are regurgitated, but any bile-staining of the vomit is of the utmost significance. In general, the lower the site of the obstruction in the bowel, the later will be the vomiting. However, it must be remembered that the lesion has been present for some time before birth in the case of atresias and meconium ileus, and the vomiting occurs in the first day of life almost invariably. If the vomiting is delayed, then it suggests that the obstruction is incomplete, as in cases of megacolon with a short aganglionic segment, or that it was not present before birth. In Hirschsprung's disease the passage of

meconium is delayed and the stool is small and hard, or in narrow toothpaste-like ribbons. Then the vomiting is delayed because the obstruction is not complete, and is low down. Four cases of Hirschsprung's disease, in this series, presented with serious symptoms during the first month of life and two died within three months of birth.

Similarly distension of the abdomen will indicate that the vomiting is not innocent. On the other hand, it is not always found. It may be localized to the epigastrium when the obstruction is in the duodenum, or generalized. In the latter case the abdomen very rapidly becomes tense and shiny. The skin over it takes on a rather dusky hue and shows dilated veins. The appearance of the distension is quite different in obstructions of the ileum, when it is central, from that occurring in obstructions of the lower colon when the distension, which is more gross, is noticed in the flanks and is accompanied by a flaring of the costal margins.

Meconium is generally passed during the first few hours after birth. It begins to change and show the presence of milk curds in seventy-two to ninety-six hours. Any delay or any alteration in the initial appearance from the normal black, tarry stickiness should be viewed with grave suspicion. Changes in colour or consistency may be associated with intestinal obstruction. It must be noted, however, that meconium is passed and is coloured to some extent even in atresias.

Delay in the passage of milk curds over four days must be viewed with suspicion. On the other hand, a changing stool in an infant with obstruction will show that the obstruction has occurred since birth or is incomplete.

Hence, a newborn baby who vomits bile, or has vomiting and abdominal distension, or has any abnormality of the meconium, should be regarded as having intestinal obstruction until proved otherwise. A plain radiograph should be taken forthwith.

Other symptoms and signs occur. Before vomiting commences the baby often refuses feeds. Blood may be seen in the vomit or in the stools. Melæna may indicate a strangulating lesion : that is, a volvulus associated with malrotation. Distension of the abdomen by restricting respiratory movement may cause considerable respiratory embarrassment in the infant, leading to cyanosis and sometimes to pulmonary collapse.

Peristalsis may be seen, and if it passes from left to right across the upper abdomen the obstruction is probably in the stomach or duodenum. Palpation of the abdomen will sometimes reveal ill-defined, firm, pitting masses in the lower abdomen. These are due to retained meconium and are found in meconium ileus and some cases of imperforate anus. A pyloric tumour will be found in the occasional cases of hypertrophic pyloric stenosis presenting just after birth.

Rectal examinations are not often helpful and, in fact, can be misleading. The collapsed microcolon gives an impression of stenosis or atresia at the recto-sigmoid junction. This fallacious finding occurred in several of the cases Spriggs (1912) described.

The mother will often have suffered from hydramnios during her pregnancy. This complication is not seen in mothers of children who develop hypertrophic pyloric stenosis. The familial tendencies in cases of neuromuscular dysfunction and meconium ileus have been observed.

Investigations.—Plain films should be taken in all suspected cases. It is preferable to take postero-anterior projections with the infant prone and then in the erect position. The latter film will show fluid levels, but if there is any reason why the infant cannot be held upright, then a lateral projection in the supine position will suffice.

Radiological examination of normal newborn infants shows that air enters the stomach immediately after birth and rapidly travels down the small bowel and is found in the colon in four hours. Any delay in the onward progression of air indicates an obstruction (*Fig.* 248).

An excessive amount of air will collect in the bowel above the obstruction and dilatation will be seen. The infant normally shows a large amount of air in the small bowel as compared with an adult. Nevertheless, any obstruction is rapidly recognized. The number of loops of distended bowel actually diminishes, but the distension of the bowel increases enormously. The shape of bowel outlines, which are normally polymorphic and ill-defined, become more clear and honeycomb-shaped. This appearance is presumably due to the individual coils pressing on each other. In addition ' hair-pin ' loops or ' step-ladder ' patterns may be seen.

Identification of the bowel involved is more difficult in the newborn than in older patients, in whom dilated jejunum shows a coiled-spring appearance (*Fig.* 255). Ileum has a smooth wall and colon is identified by its position and haustration. In the infant all dilated bowel has a smooth outline. Nevertheless, with care, it is possible to get a good idea of the level of the obstruction.

A duodenal obstruction causes dilatation of the proximal duodenum and stomach, and the amount of air in the bowels is reduced. Furthermore, if a catheter is passed and the stomach contents are aspirated, all the air can be removed, giving a useful confirmatory test for duodenal ileus (*Figs.* 249, 250).

Colonic obstructions can be differentiated from small bowel obstructions because the dilated colon shows as a gas-filled structure, of almost even bore, lying in the flanks and running across the epigastrium in one continuous sweep.

An obstructed small bowel shows as gas-filled distended loops in the centre of the abdomen. Generally there is no apparent continuity between the various loops and there is a gradual increase in distension from one loop to the next (*Fig.* 256).

If there is still doubt as to the identification of the distended bowel, a small amount of radio-opaque emulsion can be run into the colon. In small-bowel obstructions the colon will be seen as a thin thread running round the abdomen. Hirschsprung's disease, that is megacolon with an aganglionic segment in the lower colon, will be at once recognized (*Fig.* 251).

There is one pitfall to be noted. Because the microcolon, below an ileal obstruction, has never been distended, it is quite difficult sometimes for the radio-opaque medium to run, and a fallacious picture suggesting a blockage in the colon may result (*Figs.* 253, 254).

The danger of water intoxication resulting from enemata in megacolon should be remembered. Hiatt

(1951), noticing the symptoms of weakness, anorexia, and sweating, has demonstrated the dilution of serum electrolytes after enemata. It is obvious that every effort must be made to siphon back the enema fluid and in suspected cases of megacolon a limit should be fixed as to the volume of fluid introduced.

Neuhauser (1946) described the X-ray appearances in cases of meconium ileus which he considered would be diagnostic of this condition. He demonstrated that small air-bubbles could be seen apart from the outlines of the air-filled distended loops, and suggested that these bubbles had been driven on into the obstructed segment (*Fig.* 254).

Meconium ileus may be diagnosed by testing the vomit for trypsin. A rapid qualitative method is to drop a piece of exposed X-ray film into the vomit. If trypsin is present all the gelatin is digested off within an hour. In cases of meconium ileus nothing will happen apart from a little softening of the gelatin film.

Farber's test is useful to differentiate cases of intestinal obstruction in which the block has been complete for a period of months before birth, as in cases of atresia. Meconium, after staining, is examined for stratified epithelial cells. If present, they are derived from the liquor amnii, which was swallowed by the fœtus. Care must be taken not to include cells from the skin of the buttocks of the baby.

If these diagnostic measures are used, then not only is it possible to make a diagnosis early enough, but also to diagnose the obstructing agent in the majority of cases. The appropriate incisions can then be made and a certain amount of operating time can be saved.

ILLUSTRATIVE CASES

Malrotation of the Intestine (4 cases).—The X-ray picture is that of a duodenal obstruction, usually sited a little lower in the duodenum than in duodenal atresia. As these cases often develop volvulus soon after birth, blood is sometimes passed in the stool, and microscopy will show stratified epithelial cells in the meconium.

Case 1, N. H., was a full-term male baby. There was no family history of significance and he was the second child.

After 36 hours he began to vomit and he was soon bringing up bile. Meconium was delayed until the second day, and on the third day contained bright red blood.

Examination showed epigastric distension but no abdominal masses. X rays revealed distension of the stomach and duodenum with no air in the jejunum or below (*Fig.* 248).

The correct diagnosis of volvulus neonatorum was made. At operation the whole of the midgut was involved, and after untwisting, the cæcum and ascending colon lay below the stomach, freely mobile on a mesentery. The duodenum was examined and no other obstructing lesion was found and the child made a good recovery.

Case 2, A. E., was a first-born, full-term male child. There was no family history of similar illness and the pregnancy had been normal.

Bile-stained vomiting began on the fifth day. The meconium had been passed normally and was changing on the fifth day but did not contain blood. Gastric peristalsis was seen and X rays showed distension of the stomach, with only a small amount of air in the intestines.

It was obvious that this duodenal obstruction had been incomplete for the first four days of life and that the diagnosis was probably that of malrotation with volvulus.

This was confirmed at operation. The midgut was easily untwisted through 360° and the colon was left lying on the left side of the abdomen. There were no bands across the duodenum. The infant made a good recovery.

Case 3, M. T., was a full-term female child. Her mother had been well during the pregnancy and there was no significant family history. She was admitted to hospital on the twelfth day. Vomiting had been present

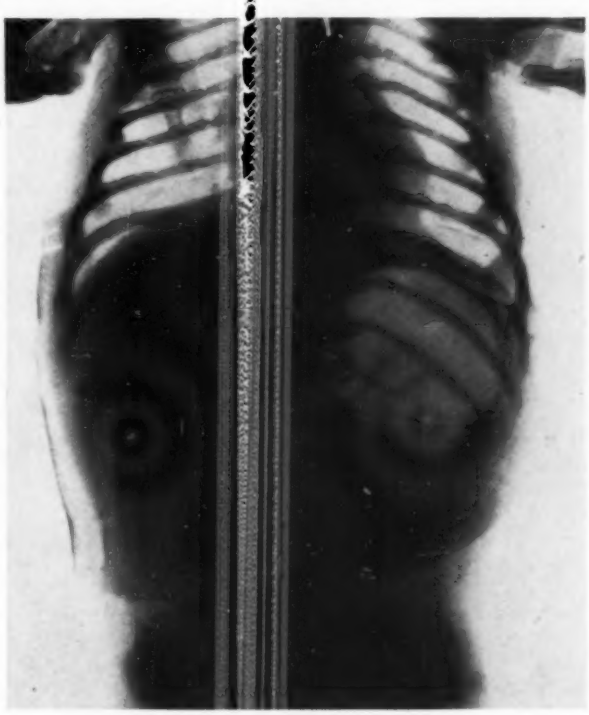

FIG. 248.—*Case* 1. Plain film, taken three days after birth, showing distension of stomach and first part of duodenum, and no air in the intestines. Definite hold-up in second part of duodenum due to volvulus.

from the fourth day and meconium had been passed normally, followed by changing stools. She had been completely constipated from the eighth day.

A barium meal, done at another hospital, showed a complete obstruction of the third part of the duodenum. As in the last case, a diagnosis of volvulus was made and confirmed. A band of peritoneum was found lying across the duodenum and had to be divided. The colon was pushed over to the left side of the abdomen.

This child made a very satisfactory recovery, until the eighth post-operative day. On the tenth day a laparotomy revealed recent plastic adhesions. They were divided but death resulted two days later.

Case 4, D. P., was the third child of rather elderly parents. There was no significant family history and no history of illness during gestation. Admission to hospital occurred on the twenty-first day because of vomiting and loss of weight. Although the child had an imperforate anus and rectovaginal fistula, meconium had been passed quite normally per vaginam.

However, on examination, there was impaction of fæces in the lower colon and it was believed that this was the cause of the vomiting. Dilatation of the rectovaginal fistula was performed with temporary improvement only

on each occasion. An iliac colostomy was therefore made. The vomiting now improved and the child returned home for a time, but there she had attacks of vomiting lasting three to seven days.

She was re-admitted during a bad attack and a plain film gave the correct diagnosis (*Figs.* 249, 250). It showed enormous dilatation of the stomach and first and second parts of the duodenum.

At laparotomy incomplete rotation of the intestines was found. The cæcum lay just lateral to the second part of the duodenum, and peritoneal bands ran across

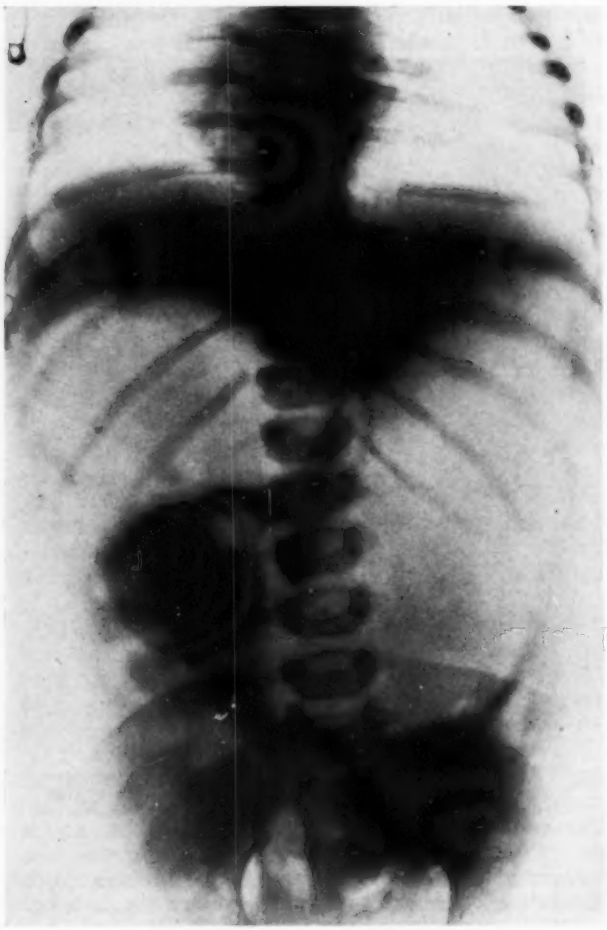

FIG. 249.—*Case* 4. Another case of obstruction in the second part of the duodenum due to malrotation. Enormous distension of first and second parts of duodenum was confirmed by aspiration of air. (*See Fig.* 250.)

the duodenum from the cæcum to the region of the origin of the superior mesenteric artery. They were causing obstruction, and the dilated proximal duodenum was ptosed and kinked over them. The bands were divided and the child has not vomited since.

Intestinal Atresia (2 cases).—Cases of duodenal atresia commence to vomit immediately after birth and X-ray examination shows that air has not progressed into the small bowel. They can be differentiated from cases of malrotation by the early onset of symptoms and the absence of squamous cells from the meconium.

Atresia of the small bowel can also be differentiated from other causes of obstruction at this level by performing Farber's test.

Case 5, G. T., a female child, was born prematurely. She weighed 5 lb. 15 oz. and began to have bile-stained vomiting immediately. Meconium was passed on the first day, but was followed by complete constipation until the fifth day, when she was admitted to hospital. Jaundice appeared on the third day and deepened. There was no relevant family history.

The abdomen was not distended, and X-ray examination showed a small amount of air in the stomach, but none in the intestines.

A diagnosis of duodenal atresia was confirmed at operation. A complete blockage was found in the second part of the duodenum below the biliary papilla. The common bile-duct was markedly dilated, and a partial

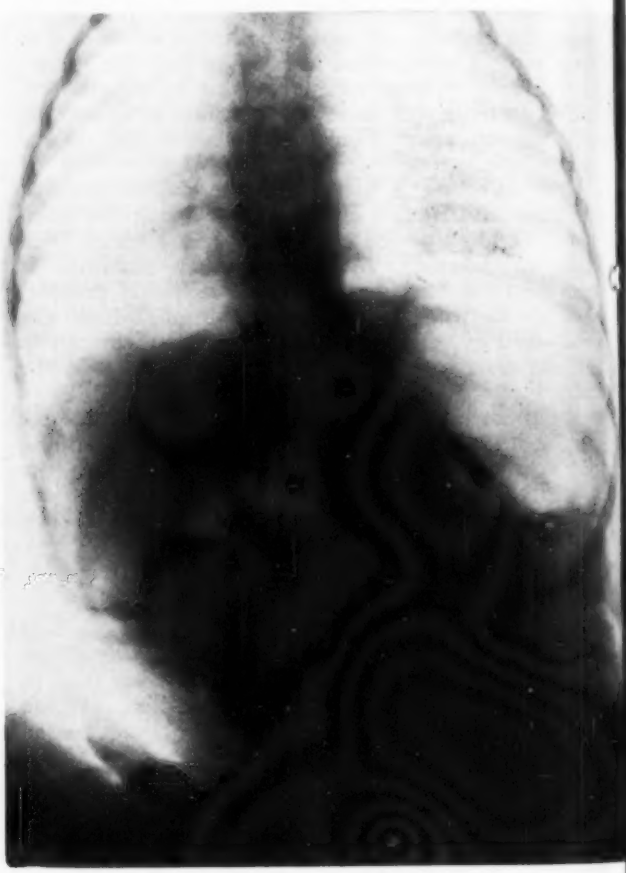

FIG. 250.—Same case. With aspiration of stomach contents, the distension of the duodenum disappears.

non-rotation of the midgut was present, the cæcum being suspended on a mesentery in the subpyloric region. A gastro-enterostomy was performed but the child died shortly afterwards.

Case 6, S. M., a female child, weighed 8 lb. 12 oz. at birth. There was no relevant family history, and one sibling was quite well. Bile-stained vomiting began two hours after birth and no meconium was passed until she was admitted to hospital on the second day. The abdomen was then distended and enlarged veins could be seen. The distended bowel was visible as a ladder-pattern, and peristalsis was observed. The distension was central and a diagnosis of small-bowel obstruction was made and a laparotomy was performed.

An atresia in the upper ileum was found. After distending the lower segment with saline, a side-to-side anastomosis was performed, but the child died forty-eight hours later.

Neuromuscular Dysfunctions (6 cases).—If the obstruction is in the colon due to a short or intermediate aganglionic segment, then diagnosis is easy. There may be a family history and the symptoms are rather slow in onset and gross distension of the abdomen with a typical colonic pattern occurs. Confirmatory evidence results if a little barium is run into the colon.

If there is agenesia of ganglion cells in the ileum then the mode of onset, the central distension, and the plain films give a picture of ileal obstruction. A family history, or the presence of cornified cells in the stool, will differentiate such a case from atresia, and the presence of trypsin in the vomit will exclude meconium ileus.

Case 7, T. R., a male child, weighed 7 lb. 11 oz. at birth. Labour had been induced for post-maturity. There was a sibling who was well, and there was no family history of any similar troubles.

Bile-stained vomiting began on the first day. Meconium was not passed for 36 hours and even then the stools were noticeably constipated. The infant was cyanosed and dehydrated. The abdomen was distended, and no masses could be palpated.

A diagnosis of intestinal obstruction was made but the infant was not investigated further. A laparotomy was performed.

The colon was dilated, hypertrophied, and increased in length as far as the rectosigmoid junction. At this level and below, it was collapsed. No procedure was undertaken and the child died next day.

Post-mortem examination confirmed the diagnosis of Hirschsprung's disease with a short aganglionic segment.

Case 8, C. B., a male infant, weighed 6 lb. 10 oz. at birth. There was no family history of any significance. Vomiting began ten days after birth and became persistent. Typical distension of colonic type was present on admission to hospital. A diagnosis of Hirschsprung's disease was made and there was some improvement with conservative treatment.

Later, however, the condition, which had been unsatisfactory, deteriorated and the child was re-admitted. X rays gave a picture consistent with a short aganglionic segment. A rectosigmoidectomy was performed but the child died seven days later.

Examination of the operation specimen confirmed the diagnosis.

Case 9, B. K., a male infant, had no relevant family history.

Vomiting began on the second day and there was delay in the passage of meconium, which was quite hard. He was admitted to hospital after three weeks, when it was found that the abdomen was very distended, and outlines of the bowel could be seen. The enormously distended colon was identifiable. A barium enema confirmed the clinical impression of Hirschsprung's disease (*Fig.* 251).

He was treated conservatively. Pneumonia supervened and he died without any surgery being performed.

Case 10, M. B., was a full-term male infant without any family history of importance. Progress was normal for three days before he began to vomit. There was no delay in the passage of meconium but it was obviously constipated. On admission the abdomen was grossly distended, especially in the flanks.

A barium enema showed marked dilatation of the colon with narrowing of the rectosigmoid. He was clearly a case of Hirschsprung's disease and continued to deteriorate. A transverse colostomy was performed fourteen days after birth. He is at present progressing satisfactorily.

Case 11, N. D., a male child, had no relevant family history. He was admitted to hospital within two hours of birth because severe abdominal distension was present, which had caused difficulty with delivery.

The abdomen was grossly distended, and there were signs of intraperitoneal fluid with central resonance. Rectal examination revealed no abnormality. Eight ounces of turbid yellow fluid were withdrawn by paracentesis, and on culture yielded a growth of *Cl. Welchii*.

Plain films showed the presence of an ileal obstruction and free gas in the abdominal cavity. Laparotomy confirmed the presence of meconium peritonitis due to a small rupture in the distended ileum. The terminal ileum and colon were collapsed, empty, and in spasm. An ileostomy at the site of the perforation was performed. In spite of specific therapy for the peritonitis the infant died two days later.

FIG. 251.—*Case* 9. Barium enema in a case of Hirschsprung's disease in the newborn. A continuous loop of grossly distended bowel in the periphery is seen. The diagnosis has been confirmed by introducing some barium.

Examination of sections of the lower ileum and colon did not reveal any ganglion cells. They were absent for about two feet above the site of the apparent obstruction.

Ganglion cells were found in the duodenum but unfortunately no sections were taken from the intervening bowel.

Case 12, M. T., was a male, second born child. There was no family history of any relevant abnormalities. Bile-stained vomiting commenced immediately after birth, and meconium was not passed.

He was admitted to hospital on the third day. Apart from slight distension, the abdomen seemed normal. Rectal examination revealed no abnormality.

X-ray examination showed dilatation of a number of coils of small bowel and no gas in the colon.

At operation the terminal 12 in. of the ileum were empty and in spasm (*Fig.* 252). An ileo-transverse anastomosis was performed but the child succumbed.

FIG. 252.—*Case* 12. Operation photograph of the terminal ileum in a case of functional obstruction. It is collapsed, empty, and in spasm, and the proximal ileum is distended. No ganglion cells were found in the spastic segment.

Microscopy confirmed the diagnosis. No ganglion cells could be found in the terminal ileum or colon.

Meconium Ileus (3 cases).—Cases of meconium ileus can be identified if the vomit is examined for pancreatic ferments. Occasionally there is a history of cystic fibrosis of the pancreas in a sibling.

FIG. 253.—*Case* 13. A film of a case of meconium ileus in the prone position. Lipiodol, injected through a catheter lying in the rectum, would not run any farther into the bowel, thus falsely suggesting an obstruction in the lower colon.

Case 13, E. M., was a full-term, female infant. She was the first-born and there was no relevant family history. The distended abdomen was noticed at birth and she commenced bile-stained vomiting forthwith. A small amount of apparently normal meconium had been passed. On admission to hospital the next day, rectal examination gave the impression of an atresia high in the rectum.

X-ray examination showed a long loop of distended bowel which may well have been colon. Lipiodol was introduced into the rectum through a catheter and the radiograph seemed to confirm the tentative diagnosis of an atresia at the rectosigmoid junction (*Fig.* 253).

It was decided to make a temporary colostomy, but at operation the diagnosis was found to be meconium ileus. The pale inspissated meconium was washed out as suggested by Hiatt (1948) but the child died 24 hours later.

FIG. 254.—*Case* 15. This film of a case of meconium ileus shows distended ileum and jejunum. On the left side of the abdomen a series of bubbles, which suggests the diagnosis of meconium ileus, is seen. Lipiodol introduced by a catheter into the colon will not run farther than the pelvic colon.

Autopsy confirmed the diagnosis of cystic fibrosis of the pancreas and no abnormality of the lower colon was present.

Case 14, G. S., was a full-term, male infant. There was no relevant family history. Bile-stained vomiting commenced immediately after birth and on admission on the fifth day the abdomen was distended. There was no satisfactory history about the passage of meconium.

The vomit failed to digest the gelatin off a piece of X-ray film. A diagnosis of meconium ileus was confirmed at operation. The hard fæces were washed out, but the child died two days later.

Case 15, D. B., was a first-born male infant. No significant family history was obtained. He commenced to vomit on the second day and distension was noticed. He was admitted to hospital on the third day, by which time no meconium had been passed.

Plain films showed numerous dilated loops of small bowel and bubbles of gas in the left iliac fossa. A barium enema was attempted but the barium would not run further than the pelvic colon (*Fig.* 254).

On a diagnosis of intestinal obstruction a laparotomy was performed. The ileum was found to be dilated except for the terminal part, which was filled with hard, white, inspissated fæces. No atresia was found. An ileotransverse anastomosis was performed, but the child died four days later without a motion being passed. A post-mortem examination was not obtained.

Miscellaneous Causes (3 cases).—

Case 16, J. B., was wrongly diagnosed for some time before the true diagnosis of internal hernia was made. At birth she weighed 6 lb. 9 oz. Immediately after birth there was a period of vomiting but it is not known whether bile was brought up at this stage. The attacks of vomiting then recurred about every two months and lasted one to two weeks, during which time the infant became intensely dehydrated. Apparently she was regarded as having cyclic vomiting. At the age of one year she began to complain of abdominal pain during the attacks.

FIG. 255.—*Case* 16. Plain film showing about three coils of distended small bowel. This film was taken when the child was 15 months old and the bowel is recognizable as jejunum because of the coiled-spring appearance.

On admission there was bile in the vomits. The abdomen was scaphoid with a localized mass in the epigastrium, which showed peristalsis.

X rays showed three coils of distended jejunum and the absence of air in the rest of the intestines (*Fig.* 255).

The diagnosis was one of recurrent obstruction in the jejunum and recurrent herniation seemed to be the only explanation.

At operation a small defect was found in the transverse mesocolon, through which was herniated a loop of jejunum. It was fairly easily reduced and the defect repaired, resulting in an uneventful recovery.

Case 17, A. G., a male child, had a birth weight of only 5 lb. 2 oz. His mother had been under treatment for syphilis for twelve months. The confinement had taken place in a maternity home and the baby, being well at birth, had been discharged home in the usual time. There was no evidence of congenital syphilis and the W.R. was negative.

He was admitted to the Children's Hospital because of bilious vomiting of twenty-four hours duration. The mother, however, had noticed distension for seven days. The abdomen was very distended and shiny. The general condition was very poor.

X-ray examination gave a typical picture of ileal obstruction.

At operation five inches of the terminal ileum were strangulated behind an adhesion lying in the R.L.Q.

FIG. 256.—*Case* 18. Many discontinuous distended loops indicative of obstruction low in the small bowel. Fluid levels are seen and also the flattened bowel walls, due to the impingement of one on the other. A hairpin loop is present.

The attachments of this adhesion were not properly identified. Above, the ileum was grossly dilated and on point of perforation at many places. Peritonitis was present and was fairly advanced.

The strangulated bowel was not viable. An extensive resection and anastomosis was performed, but the child died within twenty-four hours.

Case 18, F. T. D., a male infant, weighed 8 lb. 3 oz. at birth. His mother had been well and it was her second pregnancy. The child had commenced to vomit on the first day, but bile was not returned until the third day. Meconium was passed normally, and was beginning to change. He was admitted to hospital on the fourth day and plain films of the abdomen showed marked dilatation of a few coils in the upper abdomen, with fluid levels (*Fig.* 256). These enabled a diagnosis of obstruction of the upper small bowel to be made. A more accurate diagnosis was not attempted before operation was undertaken.

A loop of ileum was found to be adherent to the parietal peritoneum in the left lower quadrant. There

was kinking of the bowel sufficient to cause a non-strangulated obstruction. The bowel was easily freed from its point of adherence. As the bowel wall at this point appeared inflamed and œdematous it was decided to perform a side-to-side entero-anastomosis to short-circuit the unhealthy section.

Progress was very satisfactory until six days after the operation, when the abdomen became distended and the

FIG. 257.—*Case* 18. A picture of gas under the diaphragm. This is the appearance often seen in meconium peritonitis, resulting from a perforation proximal to an obstruction.

child commenced to vomit again. X rays showed free gas in the peritoneal cavity (*Fig.* 257). Laparotomy revealed that a small leakage had occurred from the suture line. It was repaired, but in spite of antibiotic therapy the infant died from peritonitis.

DIFFERENTIAL DIAGNOSIS

Various other conditions of the newborn may simulate intestinal obstruction to some extent, but rarely give any real difficulty in diagnosis. Such anomalies include œsophageal atresia and imperforate anus. Although the former does cause vomiting, the inability to swallow saliva and the attacks of cyanosis with feeding are characteristic. The failure of a catheter to pass through the œsophagus into the stomach will be diagnostic.

An imperforate anus will cause all the symptoms and signs of intestinal obstruction, but these are generally delayed for a day or more. The diagnosis should be made by inspection of the anus at birth.

Cases of hypertrophic pyloric stenosis are generally symptomless for two to three weeks after birth. Rarely the vomiting begins at birth, but the vomit is not bile-stained and the pyloric tumour may be palpated. Pylorospasm may cause symptoms within a day or two of birth, but the vomiting is intermittent and is not accompanied by distension or interference with the passage of meconium and stools. Cases of hiatus hernia and chalasia of the lower œsophagus also do not have any interference with bowel function and in neither of these conditions nor in pylorospasm is bile regurgitated.

Parenteral causes will occasionally cause difficulty with the diagnosis. Raised intracranial pressure and septicæmia are important causes of persistent vomiting in the newborn.

SUMMARY

The importance of early diagnosis of neonatal intestinal obstruction is emphasized.

It is imperative to have taken plain films of the abdomen in any suspicious cases. They are of great value in making an early diagnosis. Further it is suggested that by using the various diagnostic measures available it is possible to make a pre-operative diagnosis of the cause in most cases.

The causes are discussed in eighteen cases of neonatal intestinal obstruction.

I wish to thank the surgeons of the Royal Manchester Children's Hospital for allowing me to report their cases and also Professor W. F. Gaisford for his helpful criticism and advice.

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SIMPLE INFLAMMATORY STRICTURES OF THE SMALL INTESTINE

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In the past 20 years much has been written about chronic stenosing lesions of the small intestine, mainly under the title of regional ileitis or Crohn's disease, but there is still a great deal which is uncertain. The two cases reported here were seen over a period of eighteen months and are considered of interest in view of their marked similarity, clinical and pathological, and in view of their uncertain aetiology.

Intrinsic strictures of the small intestine are seen from time to time by most general surgeons but their infrequency, and still more the difficulty of small intestinal investigation, combine to make their diagnosis difficult before acute obstruction supervenes. This applied with special emphasis to both these cases aged 71 years. It was not unnatural that in each case carcinoma should be strongly suspected, particularly in view of the age and loss of weight.

FIG. 258.—*Case* 1. Photograph of resected ileum demonstraing long tubular stricture with ulcerated mucosa.

After exclusion of this by means of repeated radiographs one was for a time labelled as functional, but in each case recurrent acute exacerbations of pain made laparotomy imperative.

CASE REPORTS

Case 1.—W. H., aged 71 years, gave a history of 4 or 5 attacks of gripeing central abdominal pain during the 6 weeks prior to admission. After the pain had been present for several hours he would start to vomit and after about 3 vomits he would feel easier, the pain going. During the attacks his appetite was very poor but in between it was normal. He had been losing weight for some time, about 2 st. in all. There had been no bowel disturbance. He had a cough without sputum which was aggravated and possibly caused by a pharyngeal pouch which was known to have been present for 50 years. This caused difficulty with his food but not sufficient to cause him to seek medical advice. It was a familial characteristic, since his mother, brother, and sister had a similar condition.

There was nil relevant in his previous medical history and he had always been healthy.

ON EXAMINATION.—Showed a thin, highly coloured man. His hæmoglobin was 112 per cent. He had a mild degree of emphysema confirmed by chest radiograph which also showed evidence of old fibrotic tuberculosis at the right apex. His heart was fibrillating. His abdomen revealed nothing abnormal apart from vague tenderness in the right iliac fossa.

INVESTIGATIONS.—Barium meal revealed a pharyngeal pouch but no further abnormality on follow-through. Barium enema and sigmoidoscopy showed a normal large intestine.

Ten days after admission his abdomen became distended and visible peristalsis was noted making laparotomy imperative.

AT OPERATION.—Laparotomy, under general anæsthetic. A right paramedian incision was made. Many loops of distended small intestine were seen which ended about 4–5 ft. from the ileocæcal angle in a narrow, thickened length of bowel 6 in. long (*Fig.* 258). No other abnormality was found in the abdomen. The stricture was resected and an end-to-end anastomosis performed.

Following operation the patient made an uneventful convalescence, remaining in good health for 9 months,

FIG. 259.—*Case* 1. Section shows normal bowel wall passing into the area of the stricture. In this latter part ulcerated mucosa and gross fibrosis of the mucosa can be seen. (H. & E.) (× 3.)

dying suddenly from a coronary thrombosis. A post-mortem was performed confirming the cause of death and finding no other disease.

PATHOLOGICAL REPORT.—The specimen consists of 17 cm. of ileum. The proximal 6 cm. are dilated, but show no other abnormality. The distal 1 cm. appears to be normal. The intervening 10 cm. show thickening of the wall and decrease in size of the lumen. The mucosa is covered in places by a whitish exudate, and the normal transverse mucosal folds are absent. The subserous lymphatics are distended and appear as beaded white lines. Twelve lymph-nodes were present in the attached mesentery.

Sections of the thickened area of ileum show superficial ulceration of the mucosa which seldom extends through the muscularis mucosæ (*Fig.* 259). There is marked increase in the amount of fibrous tissue present in the submucosa and the arteries show some degree of obliterative endarteritis. There is no diffuse cellular proliferation and no focal proliferation of the lymphoid tissue. In some areas the fibrous tissue extends into the muscle coat. There is some subserosal fibrosis but it is much less than in the submucosa. No tubercle follicles can be seen in any of the coats of the bowel wall and the dilated lymphatics seen by the naked eye have collapsed and are not visible in the sections.

Sections of all the lymph-nodes show a marked degree of catarrhal adenitis only. There are no tubercle follicles and no other specific changes.

Case 2.—W. G., aged 71 years, had led a healthy life until 7 months prior to admission when he began to experience abdominal pain. Recently this had become colicky in nature and culminated in a severe attack of intestinal colic as the result of intestinal obstruction. There was no vomiting and his appetite was good. Some months before admission he had had a few attacks of loose motions with intervening periods of constipation. He had lost 2 st. in weight in 8 months. He had never had a cough and always been healthy.

ON EXAMINATION.—A pale, thin man who had obviously lost weight. His lungs and heart were normal for his age. His abdomen was soft and not distended, with no abnormal masses palpable. There was no tenderness.

INVESTIGATIONS.—On more than one occasion at another hospital a barium meal and enema had been performed with negative findings. Radiograph of his chest was normal. His hæmoglobin was 106 per cent.

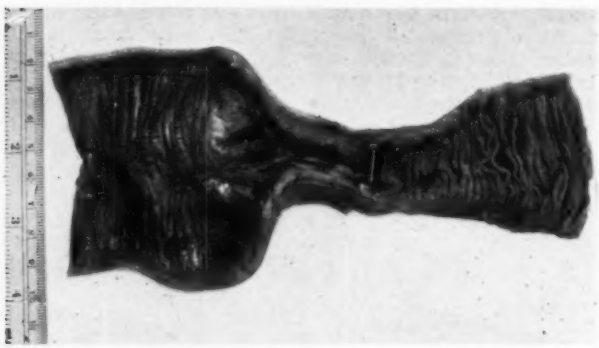

FIG. 260.—*Case* 2. Photograph of resected upper ileum demonstrating stricture with greatly thickened bowel wall and proximal dilated intestine as the result of obstruction.

Before further investigations could be performed he suddenly developed a violent attack of central abdominal pain, colicky in nature. He vomited, and a mass became palpable to the right of the umbilicus. His pain settled in a few hours but subsequent review after operation 18 hours later suggested that an intussusception had occurred and later reduced itself.

AT OPERATION.—Laparotomy, under general anæsthesia. A right paramedian incision was made and free fluid was found in the peritoneal cavity. Coils of dilated intestine were found which ended in a stricture 2½ in. long with thickened walls and showing evidence of intense congestion (*Fig.* 260). The stricture was situated about 4 ft. from the ileocæcal angle. No other abnormality was found in the abdomen and the diseased bowel was resected. A side-to-side anastomosis was performed in view of the inequality of bowel ends.

The patient made a satisfactory and uneventful convalescence.

PATHOLOGICAL REPORT.—The specimen consists of 58 cm. of small gut with the mesentery; 11 cm. from the distal end the bowel wall is thickened for a length of 6 cm. Both the mucosa and the serosa of this segment are somewhat congested and the lumen considerably constricted. The bowel proximal to the constriction is distended, and its mucosa is reddened; 17 lymph-nodes are found in the mesentery.

Section through the constricted segment shows much of the mucosa to be destroyed and replaced by granulation tissue heavily infiltrated with polymorphonuclear leucocytes and less numerous plasma cells and lymphocytes. The thickening of the wall is mainly due to fibrosis and patchy lymphocytic and plasma-cell infiltration of the submucosa and inner zone of the muscle coat (*Fig.* 261). The muscularis mucosæ is hyperplastic, its fibres extending throughout the fibrotic submucosa. There is also some congestion and similar cellular infiltration of the subserosa. No tubercle follicles are present. Sections of the 17 lymph-nodes show moderate enlargement due to catarrhal adenitis only.

Comment: The appearances are those of a non-specific inflammatory process. The changes do not resemble those of Crohn's disease and there is no evidence of malignancy.

DISCUSSION

It has already been stated in the introduction that intrinsic strictures of the small intestine are uncommon, but probably the commonest cause is that process which is labelled Crohn's disease or regional ileitis. It is important therefore to discuss the relationship between this disease and the two cases reported here, especially in view of certain similarities. First described in detail in 1932 as a disease of the terminal ileum, various features have since had to be modified in the light of further experience. It is, for example, known that the disease affects not only the terminal ileum but also the colon and proximal small bowel. Brown and Pemberton (1937) pointed out that cases could be divided into two categories: (1) Involvement of a shorter or localized segment of the ileum, a single lesion; (2) A similar process which involves longer segments, usually multiple with a greater tendency to progressive extension. It is with the former type that we are concerned here and which according to Crohn may and does progress to stenotic fibrosis, remains localized, and may lead to intestinal obstruction.

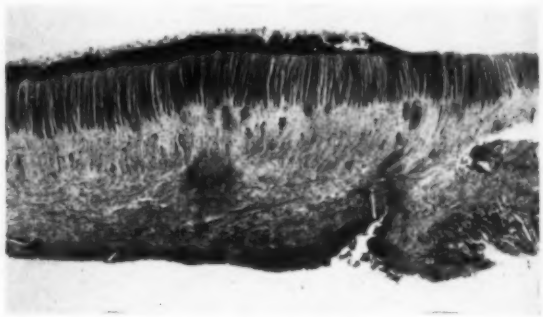

FIG. 261.—*Case* 2. Section of the stricture showing ulcerated mucosa and gross fibrosis of the submucosa with no involvement of the muscle coats. (H. & E.) (× 3·75.)

By all writers the terminal ileum is recognized as the most commonly involved site, but reports of other parts of the ileum being primarily and locally affected and not by extension are not by any means unknown. In 21 cases reported by Bockus (1946), 12 affected the ileum only, of which 2 were regional and not terminal. As far as the present cases are concerned the site of the lesion in the upper ileum is compatible with a localized type of Crohn's disease although of an uncommon type.

The age-group most commonly affected in Crohn's disease is predominantly 20 to 40 years, 75 per cent occurring before the latter age. The advanced age of these two cases is therefore interesting. According to Bockus any age may be affected but old age is uncommon.

In reporting his experiences with 222 cases of regional ileitis, Crohn (1949) states that the diagnosis is essentially a clinical one based upon the characteristic subjective symptoms and the physical findings. The histological section, he states, shows varying degrees of acute, subacute, and chronic inflammation but are entirely non-specific. Hadfield (Blackburn, Hadfield, and Hunt, 1939), on the other hand, studied a series of 20 cases by serial section derived from various stages of the disease. He laid stress on the early histological changes and described endothelial cell proliferation in the submucous lymph follicles. In the centre of these at an early stage appear giant cells, the whole forming giant cell systems which he speaks of as specific for Crohn's disease. The degree to which these lymphoid changes are to be found varies, becoming obscured at a later stage by a diffuse non-specific inflammatory process as the result of ulceration of the mucosa. Increasing fibrosis follows and is a characteristic process of the disease. According to Hadfield the regional lymph-nodes show similar characteristic giant cell systems. A careful examination of all glands available may be required to detect them, since irrespective of size not all glands contain them, some containing many, others none at all. Examination of the sections from both cases described here reveals a non-specific inflammation extending from the ulcerated mucosa into the muscle with a marked degree of fibrosis particularly in the submucosa. There is no evidence of giant cell systems which if they were ever present must have become obliterated.

The bowel wall by itself might be considered by some pathologists to represent a late and healing stage of Crohn's disease, but examination of the lymph-nodes does not show what would be expected from the investigations of Hadfield. They show only sinus catarrh and non-specific lymphadenitis with no evidence of giant cell systems and this in spite of an adequate number of glands available for section in each case, 12 in one and 17 in the other.

The clinical feature of these two cases may be described as one of obstruction with a period of vague prodromal symptoms of uncertain origin in no way akin to the classical picture of regional ileitis with diarrhœa, abdominal pain, loss of weight, and anæmia, etc., occurring usually over a period of from months to years. Cases labelled as Crohn's disease have, however, been described on several occasions with only a brief history and with obstruction as the initial sign. In Crohn's series there were two such cases but no specific details of pathology are given, while Bockus mentions 3 out of 21 cases of a similar nature.

From the foregoing comparison of age, site of disease, symptoms, and pathology it becomes apparent that many features manifest in these 2 cases have been described in others labelled as Crohn's disease, although taken together they form a most uncommon type. From the pathological standpoint the present cases resemble to the naked eye and in some microscopical respects Crohn's disease but there are none of the specific giant cell systems either in the bowel or more particularly in the lymph-nodes. If these are the features on which chronic Crohn's disease is diagnosed then these cases cannot be classified under this heading. Still less do they represent the acute stage of regional ileitis in which no giant cell systems are found and may or may not progress to the chronic type.

As with regional ileitis, it is important to exclude the possibility of tuberculosis. This is commonly stated to affect the intestine in two ways, as an ulcerative and a hyperplastic process. In the former, pulmonary tuberculosis is almost invariably associated and on healing multiple stricture formation is not uncommon. Although no examinations were made for acid-fast bacilli, chest radiographs of both patients showed no active disease. In *Case* 1, however, the chest radiograph revealed evidence of old fibrotic tubercle at the right apex. There was a history of chest trouble thirty-three years previously but this did not require hospitalization and was not regarded as serious by the patient. Post-mortem examination of this patient confirmed the absence of active disease. There are few cases reported in the literature of stenosis due to healing of tuberculous ulceration. Four cases were described by Milroy Paul (1945) showing the typical narrow annular stricture quite unlike the two described here. The hypertrophic type, well known in the older literature and said to occur predominantly in the ileocæcal region, is now regarded with suspicion. Under this title a series of 29 cases was reported by Davis, 1933; 23 occurred in the ileocæcal region, 5 in the large intestine, and one in the jejunum. The last case in some ways resembled those reported here, but the diagnosis was based on histology with no confirmatory demonstration of tubercle bacilli. The patient presented with obstruction after a six weeks' history of abdominal pain and the type of stricture was longer than that found following ulceration. The diagnosis in the ulcerative type of the disease depends on the presence of the typical histological appearance plus the demonstration of tubercle bacilli, both of which features were lacking in the two cases described here. In addition the fibrosis is predominantly in the submucosa and does not involve the muscle, features which are unlikely in the presence of tuberculous infection causing stricture. The difficulty in the diagnosis of hypertrophic tuberculosis has already been stressed, but since no giant cell systems were found it does not appear necessary to consider the diagnosis further.

Long tubular strictures have been reported as the result of interference with the blood-supply and superficially resemble both these cases. The best known are those which result following reduction of a strangulated hernia. This type of stricture was reviewed by Barry in 1942. In these, signs of obstruction develop at a variable time from soon after operation to many years later, but in neither of these patients had there been a previous history of hernia. Wangensteen (1942) mentions that partial infarction of the bowel as in mesenteric thrombosis may also lead to stricture formation. Both patients were elderly and therefore prone to vascular disease, *Case* 1 indeed dying nine months later from coronary thrombosis, but it is difficult to produce further evidence in favour of this view in either case.

There remain for consideration a few specific causes of infection and ulceration of the small intestine such as actinomycosis, dysentery, and typhoid. These are most uncommon as causes of

stricture formation and in the latter two diseases especially stenosis is a rare event. From neither clinical nor histological points of view is there reason to incriminate any and they need no further discussion. Syphilis is a rare cause of tubular stricture formation but clinically it would appear unlikely whilst histologically it can be ruled out by the limited area of inflammation and fibrosis being confined almost entirely to the submucosa.

Non-specific ulceration of the jejunum has been described in detail by Ebeling (1933), and of the ileum by Brown and Pemberton (1936). The aetiology of this uncommon condition remains unknown but amongst the recorded cases a few have progressed to stricture formation. This has been narrow in type, similar to that found following tuberculous ulceration and quite dissimilar to the non-specific strictures recorded here.

SUMMARY

1. Two cases of non-specific inflammatory stricture of the small intestine are described.

2. Their possible aetiology is discussed in relation to other causes of inflammatory stricture. Their similarity to Crohn's disease is discussed but in neither case were specific giant cell systems found either in the bowel or lymph-glands.

I wish to express my thanks to Sir Cecil Wakeley and Mr. E. G. Muir for their permission to report these two cases. Also to Dr. G. F. M. Hall for the pathological reports and Professor H. A. Magnus for his helpful suggestions.

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MODERN TRENDS IN BILIARY SURGERY

BY R. J. McNEILL LOVE

OPERATIONS FOR GALL-STONES

Cholecystostomy.—Drainage of the gall-bladder with removal of stones still has a place in biliary surgery. A gall-bladder which is contracted and buried in adhesions, especially if the patient is fat or frail, is wisely treated by removal of calculi, application of diathermy to the mucosa, and drainage of the viscus.

Cholecysto-enterostomy (or cholecystoduodenostomy).—This may be a life-saving operation for an impacted stone in the common duct when the condition of the patient prohibits any prolonged surgical intervention (e.g., choledochostomy). Provided that the gall-bladder is not contracted, this procedure is easy to perform and imposes the minimum of strain on a jaundiced and enfeebled patient, who might not survive even the decompression operation (*see below*). In such cases the risk of sudden lowering of the intrahepatic pressure by simple anastomosis must be balanced against the more prolonged, but otherwise safer, operation of decompression. If the patient survives the operation the stone remains dormant in the duct. I have performed this operation on 9 patients with 3 deaths, two of which were due to the hepatico-renal syndrome and the other died an hour or so after the operation, probably as a result of poisoning by pentothal, which was given in error, and the damaged liver was incapable of detoxicating the drug. The remaining 6 cases made excellent recoveries with no further biliary symptoms. Included in these is a male patient who just survived the operation at the age of seventy-nine but who wrote to me on his ninetieth birthday to say that he was quite well except that he had to get up once nightly in order to pass his water!

Partial Cholecystectomy.—This operation consists in emptying the gall-bladder, excising the bulk of the organ with scissors or diathermy knife, and coagulating the remaining portion so as to destroy the mucosa. This procedure is occasionally useful if the gall-bladder is difficult of access, especially if it tends to be buried in the liver. Also in cases of acute cholecystitis with partial gangrene of the gall-bladder, removal of the free portion with coagulation of the part adherent to the liver is the method of choice (Wright, 1946).

Cholecystectomy.—In skilled hands the average mortality of the standard cholecystectomy is about 2 per cent. Death is usually due to one of two causes—pulmonary embolism or chest complications. Both these conditions are encouraged by diminished movements of the diaphragm. In the standard cholecystectomy drainage is usually necessary and the puddle of bile and blood which collects beneath the diaphragm encourages spasm of the muscle, and respiratory excursions are also hampered by the drainage-tube. If diathermy dissection is performed, irritation of the diaphragm is minimal, drainage is unnecessary, and the patient can breathe deeply as soon as consciousness returns. Convalescence usually proceeds smoothly, and minor, but tiresome, chest complications are uncommon. In addition, the patient is spared the disturbance of the periodical removal of bile-soaked dressings, with a consequent saving of time and energy of the nursing staff.

Removal of the gall-bladder by diathermy is a simplification of Thorek's technique (Thorek, 1938) in which the gall-bladder was partly excised, the portion attached to the liver coagulated and covered with a graft of omentum or falciform ligament, and the abdomen closed without drainage. Some years ago a colleague and I (Bailey and Love, 1939) reported a series of 129 unselected cholecystectomies dealt with by Thorek's method, with 122 uneventful recoveries and no deaths. Increasing experience confirmed that, provided the gall-bladder bed is rendered dry by diathermy coagulation, omental or other grafts are unnecessary and the following procedure is safe and simple: after identification and ligation of the cystic duct and artery the peritoneum on either side of the gall-bladder is divided with a diathermy knife one third of an inch or so from its reflection on the liver, so as to leave a narrow flap on either side (*Fig.* 262). The gall-bladder is dissected in stages from its bed with the diathermy knife and any area from which bile or blood oozes is coagulated with a diathermy button. At intervals, as the gall-bladder is being dissected from the liver, the peritoneal flaps are approximated with a continuous suture so as to cover the denuded area (*Fig.* 263). After a final inspection the abdominal wound is closed. I have followed this procedure in over 400 cases, with a mortality in the region of 0·5 per cent. Two points in connexion with the operation require special attention—the risk of pulmonary infarction or embolism, and intraperitoneal chemotherapy.

Pulmonary Embolism.—Precautions such as the maintenance of fluid balance, gentleness at operation, and breathing exercises are commonly taken to avoid the tragedy of a pulmonary embolus, but there is one very simple precaution which, strangely enough, is ignored in many centres. Thrombosis originates in the veins of the calf or even of the feet, and if these veins are compressed by the weight of the leg, both local and distal venous circulation is retarded and the likelihood of thrombosis is thereby encouraged. *It should be an inviolable rule that during any operation on a patient who might develop thrombosis the heels must be supported so that the calves are raised from the table.* This is readily accomplished by means of a sandbag, a folded pillow, or, better still, by a special support of sorbo rubber which is wrapped in jaconet and bound with adhesive plaster. This support is easily prepared and the theatre staff learn to use it as a routine. Suitable dimensions are 4 × 4 × 18 in. (10 × 10 × 45 cm.) (Love, 1952). Some surgeons consider that the use of a bridge and consequent arching of the back not only causes post-operative discomfort owing to stretching of spinal ligaments, but also stretches and consequently narrows the inferior vena cava and so predisposes to distal venous stagnation. In thin patients, especially if some degree of hepatosis is present, the use of a bridge is unnecessary. However, in difficult cases the improved exposure obtained by the use of the bridge might outweigh its possible disadvantages.

If a patient subsequently develops a massive embolus, an immediate injection of papaverine may relax the associated arterial spasm and so tide him over the crisis. Therefore in every ward where cases of embolism may occur, papaverine (1 gr. or 65 mg.) in a sterile syringe should be kept in the drug cupboard, for immediate administration by the sister or nurse in charge to any patient who is seized with sudden dyspnœa and severe collapse. The syringe is conveniently kept in a cellophane

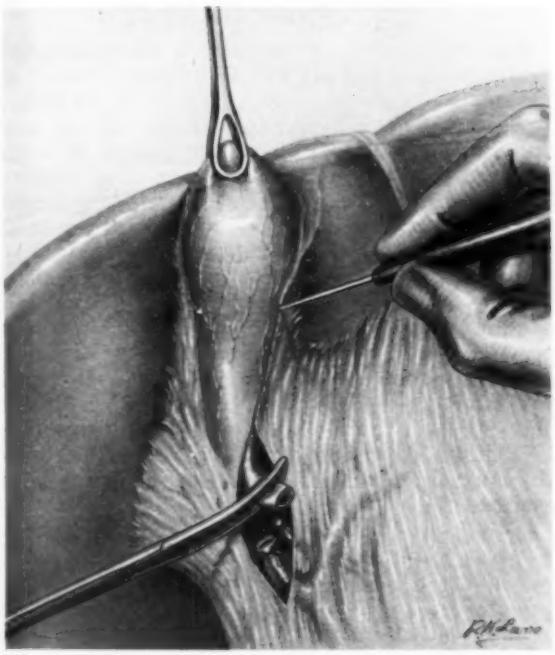

FIG. 262.—The peritoneum divided with a diathermy knife.

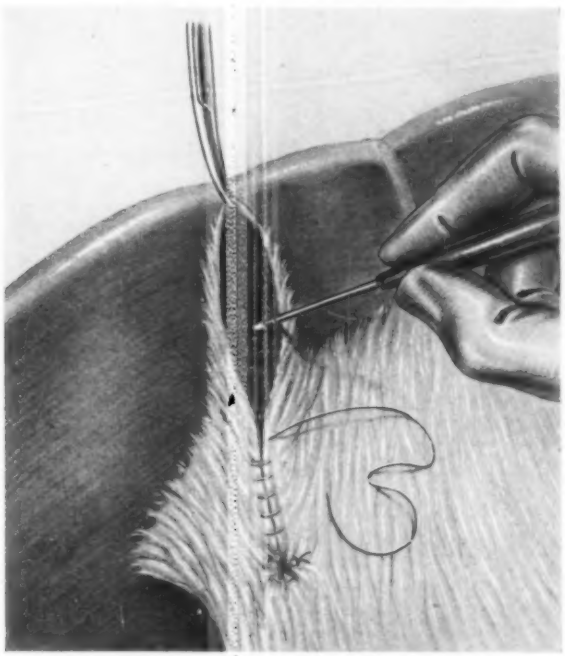

FIG. 263.—Coagulation of an oozing area in the gall-bladder bed, over which the peritoneal flaps are sutured.

envelope and plainly labelled. Thus a fatal crisis may be averted until medical advice and resuscitative measures are obtainable, and even if the diagnosis is incorrect—e.g., coronary thrombosis—no harm is done by this timely intervention.

Intraperitoneal Chemotherapy.—This precaution is advisable if recent inflammation has occurred,

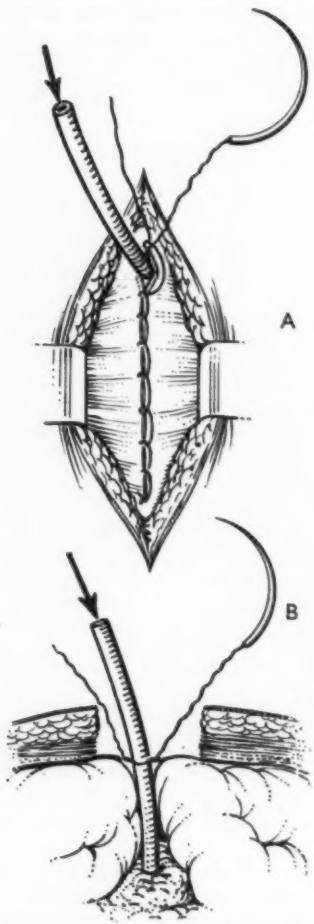

FIG. 264.—(*a*) Injection of sulphonamide suspension into the peritoneal cavity; (*b*) The suspension diffuses around the structures in the infected area. (*Reproduced from "Surgical Progress 1951" by kind permission of Messrs. Butterworth & Co.*)

or if the operation field has been soiled by infected bile. Sulphonamide powder scattered into the peritoneal cavity provokes adhesions and has the additional disadvantage that diffusion of the powder into all the recesses of the cavity is very unlikely. A very suitable preparation for local sulphonamide therapy is Mickraform. This is a 20 per cent crystalloid sulphathiazole suspension, so 5 ml. contains 1 g. of sulphathiazole. The size of the crystals is minute as compared with the ordinary sulphathiazole crystals, and therefore they cause no damage to the delicate endothelial cells. Dilution with equal parts of normal saline renders the preparation less viscous and more likely to diffuse over a wide area. In any case, if the solution is poured into the open wound a variable amount trickles out again before the suture can be completed. A simple manœuvre will prevent the waste of this unknown quantity and give the surgeon the satisfaction of knowing that the amount of drug which he wishes to use is actually retained. This manœuvre consists in the introduction of a rubber drainage-tube of small calibre down to the selected site. The peritoneum is then closed with a continuous suture, the last loop of which encircles the tube without compressing it (*Fig.* 264). The sulphathiazole suspension is then injected down the tube with any suitable syringe and, as the tube is withdrawn, the suture is tightened and tied, the wound thus being closed before any of the injected fluid can escape.

CHOLANGIOGRAPHY

For many years pre-operative cholangiography has been practised on the Continent, one of the pioneers of this procedure being P. L. Mirizzi, of Cordova, who published the results of ten years' experience in 1939. Mallet-Guy (1947), of Lyons, produced an illustrated volume recording details of 170 patients who were subjected to cholangiography and manometric readings of the pressure in the biliary passages. More recently Mallet-Guy (1952) summarizes the results of 1500 operations and states: "as long as surgery of the biliary tract is based solely on clinical data and visual and manual findings at operations, the results too often will be imperfect, and there will be a high percentage of recurrences".

I have no personal experience of manometry, which is of value in assessing the tone of the sphincter of Oddi, but five years ago (Love, 1947) I stressed the value of pre-operative cholangiography, and described a suitable technique for its performance.

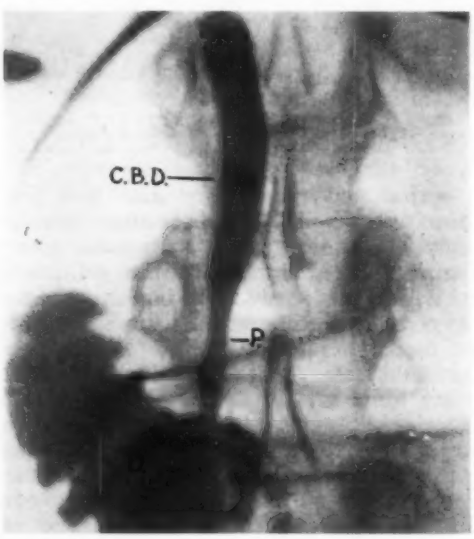

FIG. 265.—Norma cholangiogram, slight narrowing of common bile-duct at P, due to compression by pancreas, but fluid flows freely into duodenum (D).

I have availed myself of this method of investigation at 322 operations, either as a prelude to cholecystectomy, or at a subsequent operation for severe post-cholecystectomy syndromes. Owing to flaws in technique—radiological, neglect to remove an instrument the shadow of which obscured the duct, or spilling opaque fluid around the ducts—9 of the

results were uninformative. Of the remaining 313 cases, 257 showed no apparent abnormality, i.e., the calibre of the common duct was within normal limits and fluid flowed freely into the duodenum. Of the residual 56 cases the following conditions were proved or suspected: calculi 27 cases; spasm of sphincter requiring sphincterotomy 14 cases; chronic pancreatitis causing compression of duct 11 cases; carcinoma of pancreas 2 cases; papilloma of ampulla 1 case; aneurysm of the hepatic artery 1 case; (*Figs.* 265–271). It is not suggested that cholangiography

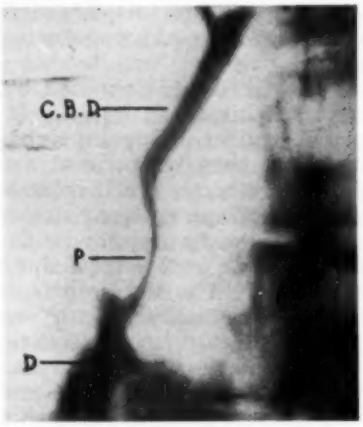

FIG. 266.—Filling defect in the common bile-duct due to papilloma (P) which was excised transduodenally. Small amount of fluid in duodenum (D).

FIG. 267.—Normal cholangiogram, showing regurgitation of fluid along duct of Wirsung (W), and free flow of fluid into duodenum (D).

FIG. 268.—Gall-stone (A) causing filling defect in common duct. No fluid enters duodenum owing to spasm of sphincter. Right and left hepatic ducts are dilated (B). The cannula used for injection is indicated by C.

FIG. 269.—Spasm of sphincter of Oddi. Dilated common duct (C.B.D.), and no fluid enters duodenum. Treated by division of sphincter with sphincterotome.

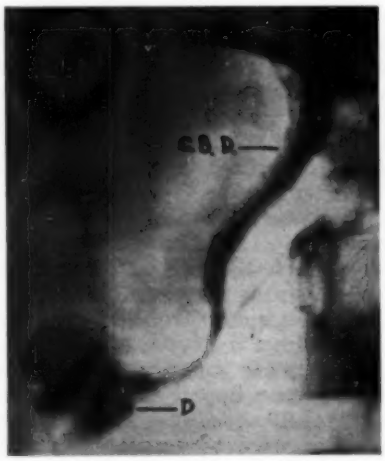

FIG. 270.—Compression of lower end of the common bile-duct due to chronic fibrosing pancreatitis.

FIG. 271.—Carcinoma of the pancreas showing gross dilatation of the common bile-duct, the typical 'amputated' appearance. No fluid enters the duodenum.

was essential for recognition of calculi in all cases. Frequently palpation would have revealed their presence, especially if performed from the *left side of the table* so that the common duct can easily be rolled between the left index finger, which is inserted through the foramen of Winslow, and the thumb. However, in some cases the distinction between a lymph-node and a calculus could only have been decided by cholangiography, which is also particularly

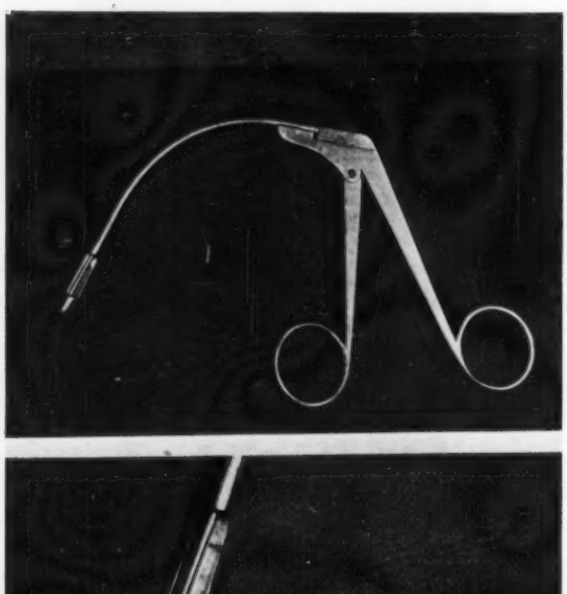

FIG. 272.—Sphincterotome for division of sphincter of Oddi. The end of the instrument with the blade exposed is shown in the lower photograph.

useful in detecting a small calculus in the impalpable lower end of the common duct.

Cholangiography serves a dual purpose—a normal cholangiogram may render exploration unnecessary in an otherwise doubtful case, and, on the other hand, the information obtained sometimes prevents the surgeon from overlooking a condition which demands exploration or some other operative intervention. I have found cholangiography of such value that I would decline to perform an elective cholecystectomy unless facilities for this investigation are available.

In this country many surgeons demur from performing cholangiography because of 'technical difficulties'. In some Continental clinics special tables are made for the purpose, but a single film, which usually provides adequate information, can easily be obtained by the following technique: On the operating-table the patient lies on a binder behind which is the casette in its appropriate position behind the gall-bladder area. The purpose of the binder is that, after the exposure is taken, two nurses or attendants, one on either side of the table, can grasp the binder and raise the patient, so as to facilitate removal of the casette. This small point is important as it obviates displacement of towels and no more than a minimal disturbance of the patient.

The gall-bladder is then exposed by the route favoured by the individual surgeon. If necessary, a distended gall-bladder is emptied so that exposure of the ducts is not thereby obscured. The liver is rotated by the right hand, which is passed between the dome and the diaphragm, so that the inferior surface then faces the surgeon, and this position is maintained by suitable forceps applied to the fundus of the gall-bladder. The three ducts are exposed and the cystic duct is nicked with sharp-pointed scissors about half an inch from its junction with the hepatic duct. Contrast medium is then injected through the aperture into the common duct by means of a malleable cannula, or, if this is not available, an efficient substitute is a suitable syringe fitted with a No. 1 hypodermic needle, to which an inch or so of a ureteric catheter is attached in order to gain flexibility. The fluid used should not be so viscous that it fails to flow freely along the biliary tree. I have found liquid neo-hydriol to be very satisfactory, but diodone gives good results. When the cannula is snugly in the cystic duct any instruments which might obscure the field are removed and the X-ray tube is manipulated into position. Some 5–10 ml. of fluid are injected during a period of about five seconds, the actual amount depending on the ease with which it flows into the biliary passages. The exposure is made as the last of the fluid is injected, and the film is ready for inspection within five minutes, during which time the surgeon can usefully be employed in removing the appendix. If the cholangiogram shows that the biliary passages require exploration, the slit in the cystic duct is extended into the common duct so that instrumentation or lavage can be performed, or if the lower end of the duct requires to be scrutinized a duodenostomy is performed. Other conditions are dealt with according to circumstances and these include spasm or fibrosis of the sphincter of Oddi. This condition deserves more attention than it usually receives, and if untreated may cause post-operative pain and dyspepsia, and varying degrees of pancreatitis (*see below*). Many surgeons are content, if the patency of the duct is doubtful, to open the duct and pass a probe into the duodenum combined with T-tube drainage in selected cases. Even then, spasm of the muscle is liable to recur, or fibrosis again contract and cause obstruction. Therefore, division of the spastic or fibrous sphincter is the best insurance against further trouble, and is performed either by means of a special sphincterotome (Doubilet and Mulholland, 1948) or under direct vision following duodenostomy. If the sphincter can be dilated, preferably with Bâkes pliable acorn-tipped probes, the sphincterotome (*Fig.* 272) is passed into the duodenum. The blade is then exposed by separation of the handles and the instrument is withdrawn until it is arrested at the ampulla of Vater. Closure of the blades effectively divides the sphincter in one plane. The three advantages of this procedure as compared with transduodenal sphincterotomy are: its simplicity, the

risk of incising the duodenal wall is eliminated, and adequate division of the sphincter is assured.

POST-CHOLECYSTECTOMY SYMPTOMS

The large majority of patients are cured or greatly benefited provided that the pre-operative diagnosis was correct and a well-planned operation executed. However, periodical check-ups over a sufficiently long period reveal the fact that in about 15 per cent of cases symptoms persist which are sufficiently severe to disable the patient; also some even declare that, as far as they are concerned, operation has been a failure. For example, in a careful follow-up Saint (1935) found that 87·2 per cent were benefited by cholecystectomy and this figure is better than those quoted by some surgeons.

Briefly, the causes of persistence or return of symptoms after a cholecystectomy are as follows:—

1. **Errors in Diagnosis.**—This is a self-evident cause and one which has diminished in numbers during the past two or three decades, owing to more careful check-up and refinements in both radiological technique and laboratory investigations. Also, it is now widely recognized that coronary disease is often associated with epigastric pain! The three surgical conditions most likely to be confused with gall-bladder trouble are hiatus hernia of the stomach, duodenal ulcer, and the ubiquitous appendix. Within the past ten years I have seen three cases of hiatus hernia who were subjected to cholecystectomy. (For some obscure reason one was much improved, although examination of the excised gall-bladder showed no obvious abnormality!) It might be mentioned that a hiatus hernia may only be discovered if the radiologist takes the precaution of examining the patient in the prone Trendelenburg position. The patient lies slightly on the right side and swallows 'up hill'. Duodenal ulceration seldom gives rise to confusion in diagnosis unless the ulcer is small and situated posteriorly. In such a case visualization will only be possible if oblique films are taken. Duodenal deformity, as distinct from ulceration, not infrequently occurs as a result of surrounding adhesions or pressure by an enlarged gall-bladder.

The variations in the clinical features of the various types of appendicitis are so well known that the surgeon is unlikely to be deceived, unless the appendix is undescended, in which case inflammation or attacks of appendicular colic may mimic gall-bladder trouble to a nicety. Mal-diagnosis in this case has no repercussion on the patient, as surgical intervention via the same route is desirable in both cases.

2. **Spasm of the Sphincter of Oddi.**—An enormous amount of experimental work has been performed in connexion with this tiny muscle. As a general rule, the tone of the sphincter diminishes after cholecystectomy, but if infection still lurks in the pancreatic or biliary passages, including the stump of the cystic duct, reflex spasm of the sphincter may occur, with attacks of epigastric pain so severe as to resemble biliary colic. The condition usually responds readily to pethidine or other antispasmodics (morphine and its derivatives are contra-indicated as they encourage spasm). Saline aperients and cholagogues, such as Dehydrocholon or Desibyl, which contain bile-salts, are also helpful as they increase the quantity of bile and diminish its viscosity. If severe attacks persist the advisability of exploration must be considered with a view to sphincterotomy.

3. **Cystic Duct Remnant.**—It is usually recommended that the duct should be ligated and divided a quarter of an inch from its junction with the hepatic duct. However, in a difficult case the surgeon might be excused if he placed the ligature even farther away from the hepatic duct, preferring to avoid the immediate hazard of possible injury to the right hepatic or common ducts in spite of the risk of subsequent cystic duct infection. Even if the cystic duct is ligated at the recognized site the small stump is in reality a diverticulum which may live up to its name as being 'a wayside house of ill fame', therefore, whenever possible, the cystic duct should be ligated flush at its junction with the hepatic duct.

Infection of the cystic diverticulum encourages or maintains infection of the common duct, with consequent attacks of cholangitis and hepatitis—which resembles the 'intermittent hepatic fever' of Charcot. Jaundice is not uncommon and may be of considerable severity during the exacerbations. The importance of the cystic duct syndrome has been emphasized by several American surgeons, e.g., Garlock and Hurwitt (1951) record 30 patients whose symptoms occurred at times varying between three months and twenty-seven years after cholecystectomy. At re-operation calculi were found in 21 patients and 24 were completely cured by excision of the diverticulum and drainage of the common duct. My personal experience is limited to 3 cases, in 2 of which calculi (one the size of a date stone) were discovered and removed with the diverticulum. In the third case an infected diverticulum (nearly 2 cm. in length) was situated behind the right hepatic duct, and was only discovered by a probe after the common duct had been opened. These cases were all cured by excision of the diverticulum.

4. **Pancreatitis.**—The association between infection of the biliary tract and acute pancreatitis has long been recognized. All stages from acute hæmorrhagic pancreatitis to chronic pancreatic deficiency are liable to occur. Recurrent subacute pancreatitis should be suspected if the patient complains of attacks of epigastric pain and abdominal tenderness, associated with nausea and perhaps vomiting. Tenderness on palpation in the left renal angle supports the diagnosis, as in this situation the tail of the pancreas is comparatively superficial. Confirmation is obtained by estimation of the serum-amylase during an exacerbation and a content approaching 100 mg. per 100 ml. is diagnostic. Tests for pancreatic deficiency are unlikely to assist, as the digestive functions of the gland are not seriously impaired in the early stages. Pethidine is prescribed during the attacks on the assumption that they are associated with spasm of the sphincter. If attacks are frequent and disabling, surgical intervention is indicated, otherwise the gland will gradually be destroyed by infection and fibrosis to the accompaniment of such severe pain and impaired health that only pancreatectomy can relieve the patient. The common duct is explored for any obvious obstruction and a cholangiogram is taken on the table. If this indicates

that the sphincter is spastic or fibrous, sphincterotomy should be performed so as to discourage further regurgitation of bile along the pancreatic duct. The common duct is then drained for some months with a T-tube, the lower end of which projects into the duodenum.

Chronic parenchymatous pancreatitis with resulting deficiency occurs not uncommonly and quite insidiously, especially in long-standing cases of infection. The patient complains of distension after food, especially fats, and epigastric discomfort. The serum-amylase is not elevated, but examination of the fæces and other pancreatic tests indicate lack of enzymes. Relief is obtained by restricting the intake of fats and replacement therapy, e.g., pancreatic preparations such as Pancrobilin or Panteric are prescribed. The patient takes a course when need arises. Symptoms gradually abate and, as infection of the biliary tract subsides, the pancreatic function improves with amelioration of symptoms.

5. Biliary Dyskinæsia.—This is an unsatisfactory diagnosis, which can only be justified when every effort has been made to exclude any of the above-mentioned causes of post-cholecystectomy symptoms. Also the mental state of the patient must be assessed, as some patients refuse to be cured.

According to Womack and Crider (1947) the post-cholecystectomy syndrome may be due to involvement of the autonomic nerves in a scar tissue around the stump of the cystic and common ducts, and they recommend resection of these nerves as a prophylactic measure during a cholecystectomy. However, most surgeons would agree with Cattell (1950), who states that " he thinks that if he made the diagnosis he would be overlooking some organic condition of the biliary tract or associated with it ".

HEPATIC DECOMPRESSION

Sudden reduction of intrahepatic pressure is apt to result in cholæmia or the hepatico-renal syndrome described by Heyd (1943). This syndrome is due to absorption of toxins from the damaged liver cells, which are deleterious to parenchymatous tissue and especially to the convoluted tubules. Sudden lowering of tension within a damaged liver, as by rapid decompression, results in hyperæmia and rapid absorption of toxins which may injure the kidney to such an extent that it ceases to function. Clinically the patient becomes drowsy, the pulse-rate steadily rises, oliguria develops and coma and anuria end the scene. Some preparations containing bile-salts are given as a cholagogue, and efforts are made to stimulate renal function by intravenous sodium sulphate (4·285 per cent) but the results are often disappointing.

The pathological changes which occur in the liver are similar to those found in the kidney when sudden emptying of the urinary bladder in a case of chronic retention is followed by renal failure.

Sudden Decompression.—If duodeno-pancreatectomy is impracticable in a case of carcinoma of the pancreas, it is customary to anastomose the gall-bladder to some part of the alimentary tract so as to relieve the physical and mental effects of obstructive jaundice. If the patient recovers from the operation he usually survives for an average of nine months or so in reasonably good health. As previously mentioned, anastomosis of the gall-bladder to the intestine is sometimes the method of choice in dealing with a stone impacted in the common duct.

A cholecysto-gastrostomy is inadvisable not only because the anastomosis of a thin-walled gall-bladder to a thick-walled stomach presents unnecessary technical difficulty, but also because the discharge of bile into the stomach stimulates rapid emptying and so induces constant hunger. Therefore, should the duodenum be sufficiently mobile a cholecysto-duodenostomy is advisable, otherwise a cholecysto-enterostomy is perfectly satisfactory. However, whatever method is used, the sudden fall in intrahepatic pressure in a case of prolonged biliary obstruction and back-pressure is a serious matter which not infrequently results in fatal cholæmia or the hepatico-renal syndrome. The mortality of any of these operations which by-pass common-duct obstruction and cause a sudden reduction of intrahepatic pressure varies with different authorities, but averages about 40 per cent. Most patients who succumb usually do so within three or four days of the operation. Sudden decompression should only be performed if the patient is unlikely to stand the slightly longer procedure of gradual decompression, i.e., when absolute minimal intervention is essential.

Gradual Decompression.—In order to prevent sudden lowering of intrahepatic pressure, I evolved the following procedure in cases of carcinoma of the pancreas when a radical excision is impracticable owing to fixity of the primary growth or to dissemination, and also for selected cases of impacted stone in the common duct. The grossly distended gall-bladder is aspirated until it is sufficiently decompressed to permit of the application of a light intestinal clamp without the risk of rupturing the viscus. Unnecessary aspiration is inadvisable as it causes a corresponding fall in intrahepatic tension. The clamp prevents further escape of bile from the puncture wound and leaves the fundus free for subsequent anastomosis. A small opening is made into the jejunum about a foot or so distal to the duodeno-jejunal flexure, through which a medium-sized rubber tube is passed proximally. A clamp is applied to the protruding end of the tube in order to prevent escape of bile during the later stages of the operation. A cholecysto-enterostomy is then performed between the fundus of the gall-bladder and the upper end of the jejunum in the following manner. The posterior seromuscular surfaces are approximated by a simple running suture, no knots being tied (*Fig.* 273). The gall-bladder and jejunum are opened and united by a posterior through-and-through running suture. The tube is manipulated so that it passes into the gall-bladder (*Fig.* 274), and it is then anchored to the anastomotic line by a single fine catgut stitch. The anterior through-and-through suture is inserted and firmly tied so that the tube is gripped by the purse-string, which prevents seepage of bile around the tube. The anterior seromuscular suture completes the anastomosis (*Fig.* 275), and is again tied in purse-string fashion so as to grip the tube. The wound is then closed except that at the lower end the tube is brought through the abdominal wall in the manner of a jejunostomy (*Fig.* 276). The tube is then

connected with a tidal drainage apparatus so that the pressure at which bile escapes is under control.

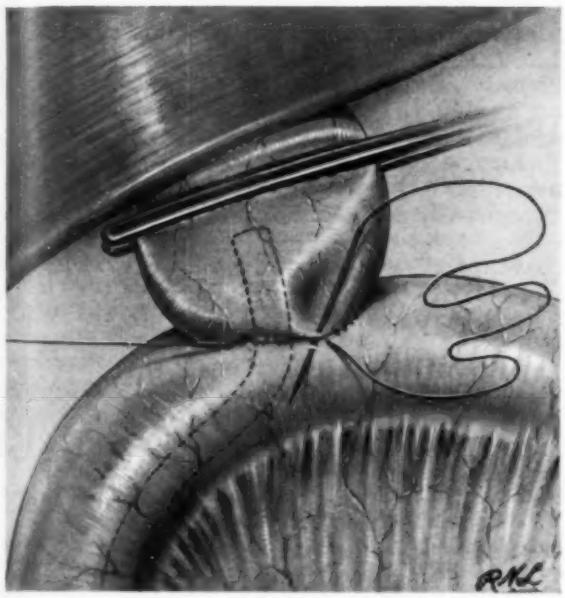

FIG. 273.—Anastomosis of the partially emptied gall-bladder to jejunum; the tube has been pushed up from a distal opening in the bowel.

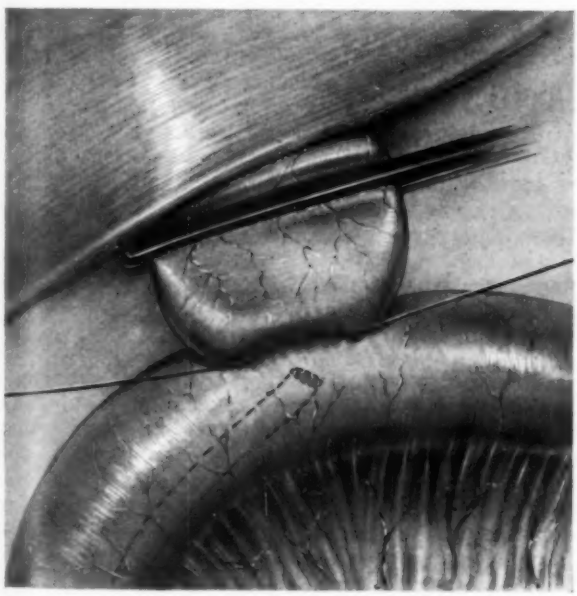

FIG. 275.—The cholecysto-jejunostomy is completed by the anterior seromuscular suture.

Cholagogues such as dehydrocholon are prescribed, and after four days or so the condition of the patient is usually much improved. Tidal drainage is then discarded and the tube is divided close to the abdominal wall so as to form an open cholecystostomy. When the tube loosens a few days later it is removed and the enterostomy closes uneventfully.

I have adopted this procedure on 9 occasions with 1 death. This was a male patient, aged seventy-nine, who had been suffering from increasing jaundice for ten weeks. He was mahogany-coloured and hepatic function tests indicated severe liver failure. Laparotomy revealed a carcinoma of the

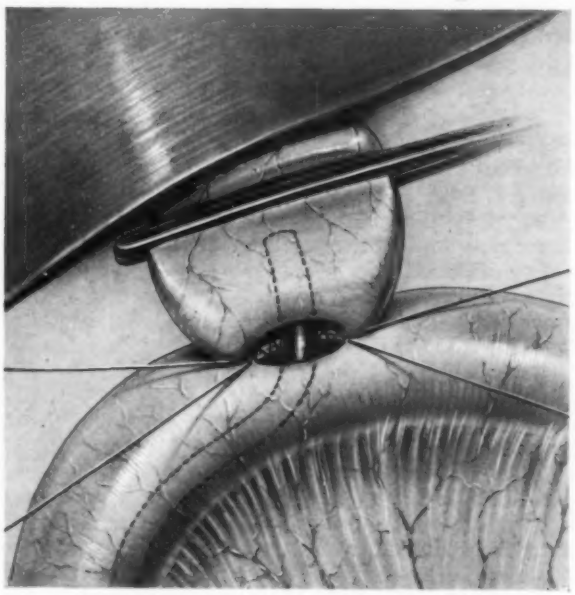

FIG. 274.—The posterior through-and-through suture has been inserted, and the tube is introduced into the gall-bladder.

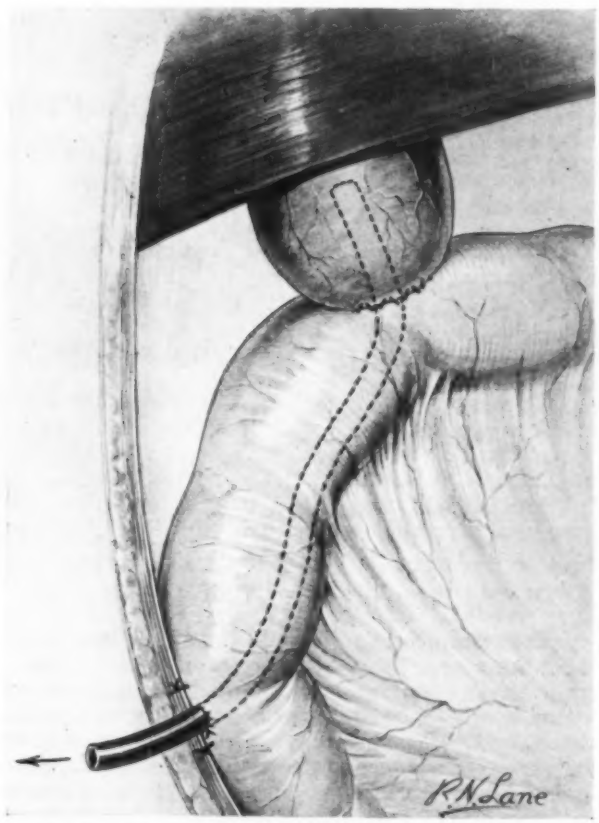

FIG. 276.—Operation completed by a jejunostomy at the lower end of the wound.

head of the pancreas, ascites, and malignant peritoneal nodules. Gradual decompression was performed, but he became increasingly drowsy, urinary output dwindled, the blood-urea rose to 280 mg., and he died in coma three days after the operation.

SUMMARY

1. A brief review is presented of the operations which are performed for gall-stones, with special reference to diathermy dissection and pulmonary embolism.

2. A simple method is described of taking a cholangiogram on the operating-table. The information so obtained may reveal some abnormality which might otherwise be missed, or, conversely, the unnecessary exploration of the common duct in a doubtful case.

3. In about 15 per cent of cases the results of cholecystectomy for gall-stones are disappointing. Some of the causes are briefly considered, with suggestions for their prevention and treatment.

4. In cases of obstructive jaundice sudden decompression of intrahepatic tension may precipitate liver failure—a condition analogous to renal failure when the urinary bladder is rapidly emptied in cases of chronic retention. A technique is described by means of which gradual decompression can be accomplished, with consequent diminution of the risk of liver failure.

I must express my indebtedness to the Radiological staffs of the Royal Northern and Metropolitan Hospitals for their co-operation in the production of cholangiograms, and to Mr. R. N. Lane for the realistic illustrations reproduced in this article.

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PULMONARY HYDATID DISEASE

BY N. R. BARRETT AND DILLWYN THOMAS

INTRODUCTION

THIS paper does not cover the possible happenings in pulmonary hydatid disease ; it describes the facets of this topic which we have seen and in it are discussed the results of the treatments we have applied. The figures include a dozen cases operated upon by Sir Clement Price Thomas.

We write again upon this subject because some of the views expressed in our earlier publications (Barrett and Thomas, 1944 ; Barrett, 1947, 1949) need modification in the light of further experience, and because the additional cases treated have provided examples of pathological states which we had not seen up till then.

DEFINITIONS

The term ' simple ' hydatid disease is used by us to indicate that the parasite itself is intact ; but a simple cyst may, upon occasion, cause suppuration in the lung. A ' complicated ' cyst is one in which the hydatid has ruptured and this accident may or may not be associated with pneumonitis. ' Primary ' hydatid disease indicates that the cyst in question has developed from an embryo derived directly from a dog. ' Secondary ' hydatid disease means that a primary cyst lodged elsewhere has ruptured and caused new cysts to develop by embolism or by direct spread. ' Daughter ' cysts are cysts which grow within the cavity left by spontaneous evacuation or inadequate surgical removal of a parasite.

PRACTICAL POINTS CONCERNING THE PATHOLOGY OF PULMONARY HYDATID DISEASE

Résumé of the Life Cycle of the Parasite.—In the British Isles it is reasonably certain that infestation takes place from dogs. It is known that, naturally and experimentally, jackals, wolves, and some domestic animals can harbour the *Tænia echinococcus*, and that hydatid cysts can grow in hogs, cats, rabbits, squirrels, etc. ; but none of these animals are important to us as carriers or hosts.

The usual life cycle of the parasite is between the dog and the sheep. The former harbours the *Tænia echinococcus*—a small tape-worm—in its alimentary canal and excretes ova in the fæces. The latter swallows the ova with its food, and, from these ova, hydatid cysts develop in its viscera. The parasite gets back into the dog when the sheep is killed and its viscera—which contain hydatid cysts—are eaten by another dog. If an ovum gets into a man instead of a sheep, the life cycle of the parasite will ultimately be broken, because the entrails of men are not eaten by dogs.

After a man has ingested ova from a dog into his alimentary canal, the embryos in the stomach burrow

through the gastric mucosa and enter a radical of the portal vein. In this way they are carried in the blood-stream to the liver where the majority settle and develop into hydatid cysts. Only those embryos which successfully pass through the capillaries in the liver reach the lungs, and the few which get through the lungs are then carried to the viscera and tissues supplied by the systemic circulation. For this reason hydatid cysts are commonest in the liver and less common in the lungs. (*Fig.* 277.)

Once an embryo has come to rest in the lung it grows into a vesicle and then into a hydatid cyst. The latter consists of a germinal membrane which secretes hydatid fluid into the cavity of the cyst and an outer protective layer of laminated hyalin. The hydatid cyst grows relatively quickly in the lung and eventually makes itself felt because it presses upon some important or sensitive structure; or because some complication occurs which affects its existence in the lung. The commonest complication is rupture of the hydatid cyst and this results in one of three possibilities. In some few cases the cyst is totally expectorated and the patient is cured. In others the tear seals off, the parasite becomes re-established and continues to grow. In the majority the liquid is expectorated but the membranes are retained in the cavity which the parasite has formed in the lung. These retained membranes can only be got rid of by the body if they can be extruded; they cannot be absorbed. Hence such a patient is liable to one of two developments. Either some parts of the germinal membrane survive in spite of complete rupture and daughter cysts grow in the space occupied by the original cyst, or the parasite dies after rupture and the laminated membrane is retained as a sequestrum which becomes infected and develops into a chronic lung abscess.

In the remarks which follow certain details in the growth and possible complications of pulmonary hydatid cysts are discussed because they have seemed important to us as surgeons.

The Significance of Multiple Cysts in Lungs.—In most cases only one parasite is found in the lungs of a patient, but sometimes two, or more, occur at the same time. If multiple hydatids (*Fig.* 278) are present the following possibilities must be considered.

The usual explanation is that at the moment of infestation the patient ingested more than one ovum, and more than one embryo succeeded in negotiating the capillaries of the liver. In such a case the liver itself may or may not be involved; but in our experience multiple lung cysts have seldom occurred in patients who have liver cysts as well. In experimental animals, such as the squirrel, it has been shown that practically all the embryos pass through the liver and take root in the lungs, so it is not very surprising to find the same thing happening on occasion in man.

Another explanation of multiple cysts occurring in the lungs is that the patient first developed a cyst in the liver, and this became complicated and shed live elements into a radical of the hepatic vein. Thus multiple 'simple' lung cysts may indicate the presence of a 'complicated' liver hydatid.

A third, and very rare, occurrence is that a hydatid growing in the wall of the right ventricle has ruptured into the cavity of the heart. Several cases of this type have been described, and the result of such a rupture has been that large numbers of cysts have grown all over both lungs.

Concerning the Size of Lung Hydatids.—It is probable that hydatid cysts flourish in the lungs because they can grow quickly. On several occasions we have observed patients in whom the cyst could only have existed for a year and yet it had attained the dimensions of a billiard ball. The smallest hydatids which can be detected radiologically in the lungs or the pleural cavity are about the size of a pea;

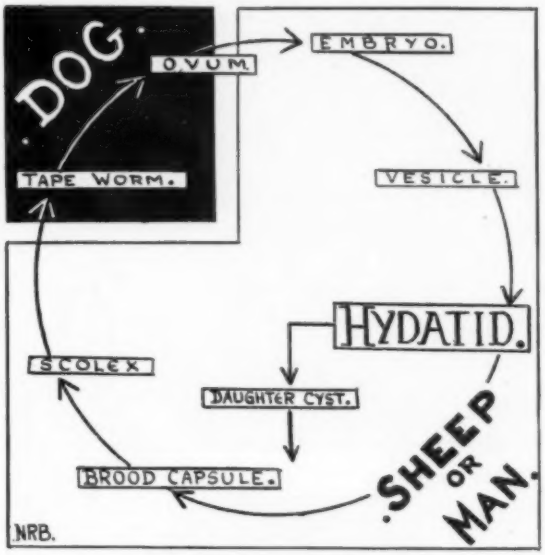
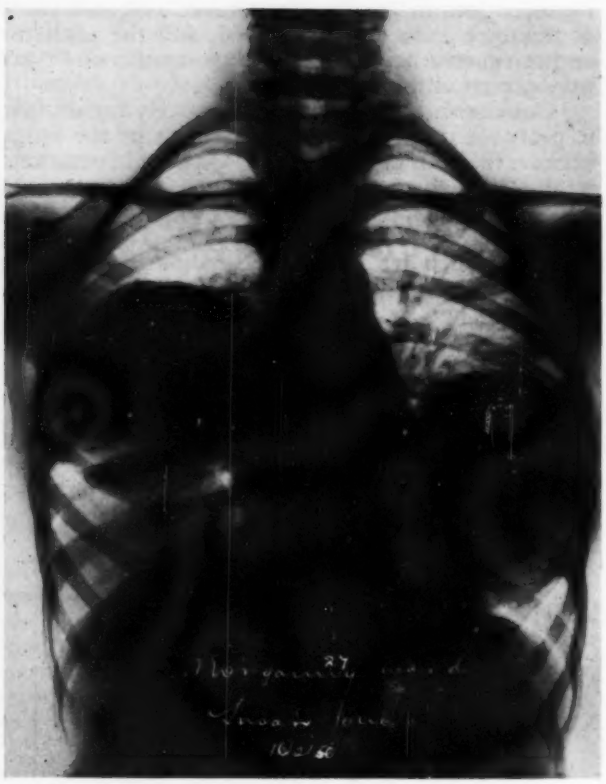


FIG. 277.—Diagram to illustrate the chief stages in the life of the parasite.

they show up distinctly in the pleural cavity if a pneumothorax is present, and in this way it has been possible to watch and measure the rate of growth. On the other hand, all hydatid cysts do not grow evenly and continuously; some appear to remain stationary for months or years and then to grow again. The largest cysts we have treated have contained about two pints of liquid; but cases have been recorded—and amongst them was the first patient described by Lænnec, a child who died as a result of bilateral giant cysts—in which the parasites have totally filled the hemithorax.

It sometimes happens that multiple cysts, *of different sizes*, are present at the same time in the lungs (*Fig.* 279) and this must cause speculation because hydatids implanted at the same time might be expected to grow at similar speeds. A possible explanation is that the patient has been infested on more than one occasion; but the usual cause is that the cysts were indeed manifestations of a simultaneous primary infestation but that a complication of some sort has befallen the smaller cyst, and has impeded its growth. A case of this type was seen recently. A girl aged 6 was found to have a simple cyst the size of an orange in the right lung and a small infected cyst in the left. In this case the infected cyst had stopped growing. Another reason for variations in size may be that the patient has harboured primary cysts in the lung and also in the liver: after some time the



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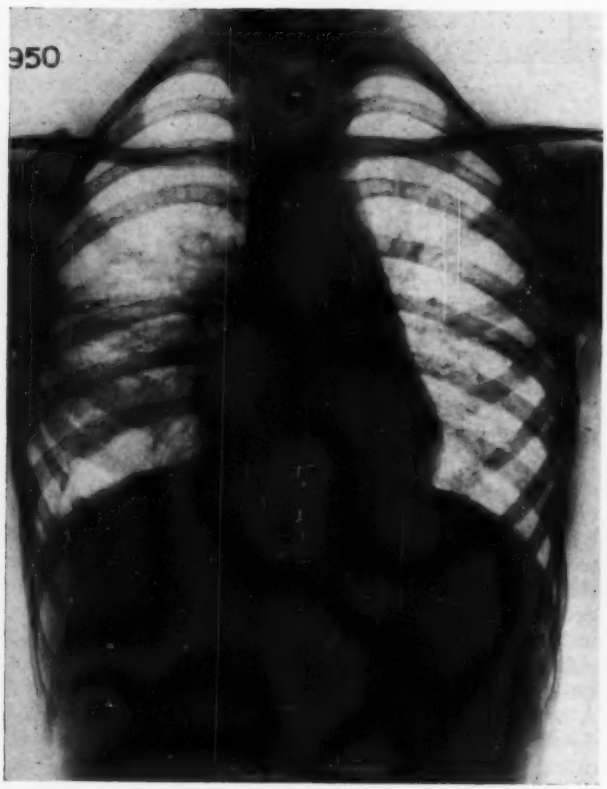


C

FIG. 278.—Radiographs of the chest of a child showing the typical appearances of multiple simple hydatid cysts in the lungs. A, Anteroposterior film; B, Lateral film; C, Appearances after enucleation of the parasites.

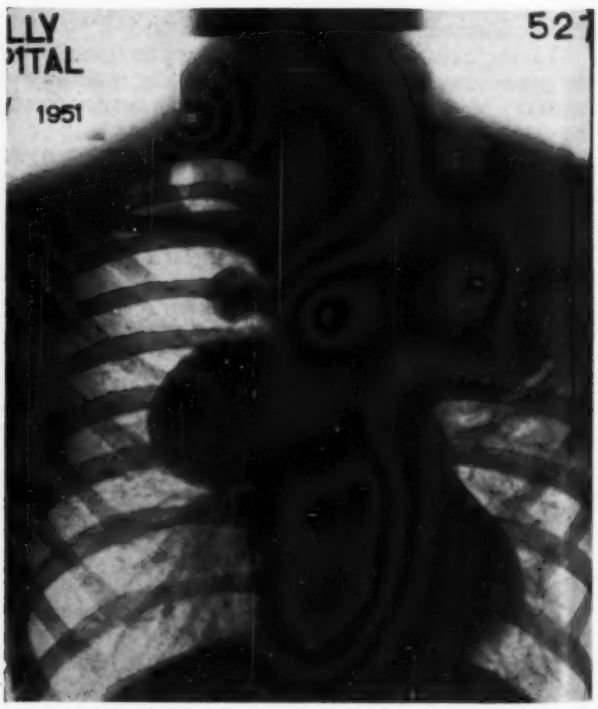


FIG. 279.—This is the radiograph of a child who harbours two hydatids in the right lung and one in the left. *Note*: The parasites are of different sizes and the one in the right upper chest is calcified, the other two were 'simple'. There were no hydatids in the liver in this case.

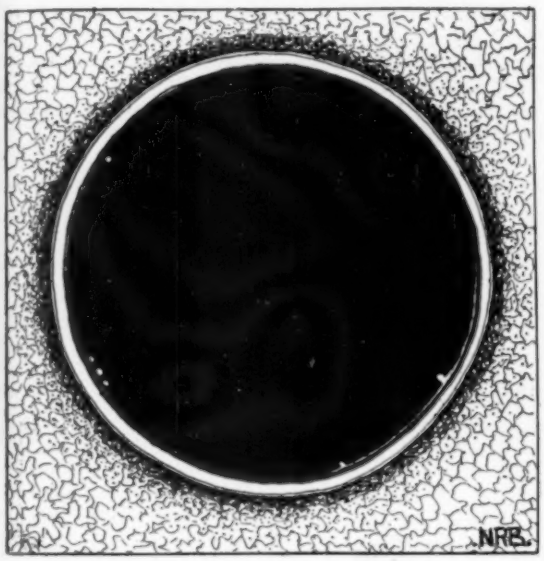
liver cyst has become complicated and produced secondary echinococcosis either by rupturing into a radical of a hepatic vein or by direct rupture into the lung through the diaphragm. Thus the presence of multiple cysts in the lungs, and particularly of cysts of different sizes, should make one suspicious that complicated hydatids may be present elsewhere in the body.

The Growth and Effects of a Simple Hydatid Cyst in the Lung.—A parasite can settle and grow in any part of the lung, and in most cases the young cyst does not abut upon a pleural surface in the first instance. It is situated deep in the substance of a lobe, but as it grows it impinges upon bronchi and other resistant structures if it advances towards the hilum, and thus there is a tendency for all lung hydatids to grow towards the surface away from the hilum. It was customary some time ago to refer to 'peripheral' and 'central' hydatid disease: the distinction was made radiologically and was used because it was believed that if a hydatid grew near the hilum there was a good chance of spontaneous evacuation into one of the large bronchi, whereas if it grew in relation to the surface bronchi the hydatid membranes could never be evacuated through the small bronchial tubes. This distinction is no longer useful because there is no case for the conservative treatment of hydatid disease based upon the chance of spontaneous evacuation of the parasite. In our experience 'hilar cysts', using the term in the manner described above, have not occurred.

Once implanted in the interstitial tissue of the lungs the embryo grows into a vesicle and then into a hydatid cyst. There is never any direct fusion between it and the lung: it fits the cavity in which it grows as a hand fits closely into a glove and at all times the laminated membrane can be completely separated from the adventitia; and thus a cyst which, at operation, cannot be immediately and completely detached from the lung has never been a hydatid.

The bulk of a hydatid cyst (*Fig.* 280) is made up of the liquid contained within it. This liquid, which has been secreted by the germinal membrane of the parasite, has been extracted from the blood of the host. It is clear, like spring water, but contains a high concentration of salt and other chemical substances. Thus the statement, which has often been made, that the laminated membrane is impermeable is not true; but it is a fact that, as yet, no drugs have been found which can reach and destroy the parasite. But even if such drugs do become available the problem of curing hydatid disease in the lung will not have been solved because the laminated membrane will be retained as a sequestrum which ultimately is certain to become infected.

Hydatid fluid contains live fragments of the laminated membrane, brood capsules, and scolices. None of these particles can easily be seen with the naked eye, but the brood capsules are sometimes referred to as 'hydatid sand'. Whether all these hydatid elements can give rise to new hydatid cysts if spilled into the tissues, or whether only the fragments of germinal membrane can do this, is of academic interest. The important thing for the surgeon to know is that hydatid fluid must never be allowed to contaminate operation wounds in the lung or the chest wall; nor must it reach the pleural cavity. In any of these sites either anaphylaxis, or new hydatids can develop. By contrast hydatid fluid in the bronchial tree does not cause bronchial dissemination of the parasites, and the reason for this is not known.



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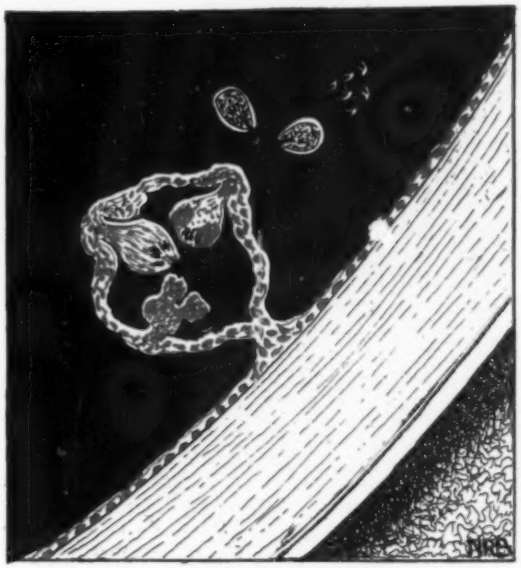


B

FIG. 280.—A, This drawing depicts the anatomy of a hydatid cyst in the lung.

Black	= Hydatid fluid	Hydatid cyst
White dots	= Brood Capsules . . . . 'Hydatid sand'	
Red	= Germinal membrane	
White	= Laminated membrane	
Stippled	= Compressed lung of host, or adventitia.	

B, Part of the wall of a hydatid cyst. Magnified.

Black	= Hydatid fluid, containing scolices and hooklets
Red	= Germinal membrane from which a brood capsule (containing three scolices) is growing
Striated	= Laminated membrane
White	= Space (potential) between parasite and host
Stippled	= Lung forming the adventitia.

If a lung hydatid grows to be large it is generally assumed that the patient may be in danger, if it bursts into the bronchial tree, of drowning in the liquid suddenly discharged into the air-passages. This accident seems to be rare and for this there are possibly two reasons. It is unusual for a cyst to rupture and to void *all* its liquid contents immediately: it is more usual for a gradual leak to occur and in such circumstances the bronchi are not flooded. Moreover the bronchial tree can re-absorb quite a large volume of liquid, so that when a cyst ruptures the patient generally coughs up much less than might be anticipated. Nevertheless, having the possibility of drowning at operation in mind, surgeons have used various types of bronchial blocker as a safeguard. These precautions are probably unnecessary, not only because modern operations succeed in removing the cyst intact, but because bronchial suction and the use of gravity drainage are adequate to deal with this emergency. (*Fig.* 281.)

FIG. 281.—Section of the lower lobe of a patient suffering from simple hydatid cyst. The patient was treated by lobectomy, but the specimen shows no irreversible changes in the lobe and enucleation would have been successful.

The laminated membrane of the parasite is white in colour and is secreted by the germinal layer inside it. It is formed by continuous deposition of a number of skins of hyalin arranged in layers like those of an onion; the oldest of these skins are on the outside. The outer skin of the laminated membrane is stretched more than the inner ones and, for this reason, when a hydatid ruptures, the membrane turns inside out immediately, and the most dangerous parts—i.e., the scolices and brood capsules—are brought into direct contact with the tissues of the host. The intact laminated membrane is strong and can often withstand the alterations in shape when a hydatid herniates out of the lung, but as soon as it has been perforated by a needle or grasped with forceps it tears. Once having started, the tear enlarges and as the liquid gushes out the laminated membrane separates completely from the tissues of the host.

The young parasite causes no permanent changes in the surrounding lung and its capsule in the host is at first composed entirely of atelectatic alveoli. As the cyst enlarges fibrous tissue begins to replace the collapsed marginal zone so that eventually a true capsule is formed. The capsule, which is always a part of the host, is called the adventitia, and it is only around really large, or long-standing, cysts that it is formed of fibrous tissue. After enucleating a hydatid it is gratifying to note that the scar generally 'melts away' in the post-operative period so that often, within a few weeks, no trace of it can be seen in the radiographs. An adventitia which has become fibrous is itself sharply demarcated from the normal surrounding lung, unless secondary infection has occurred, and thus a surgeon can easily dissect out the fibrous capsule of the host and he will find that it is seldom more than a few millimetres in thickness.

The lung is also a favourable place for the parasite because it can accommodate a space-occupying lesion simply by becoming atelectatic in the immediate vicinity. Moreover the parasite does little damage to the lung at first because it excretes no harmful substances and because, as it enlarges, it does not invade and destroy adjacent bronchi, as would a malignant tumour or a tuberculous process. The hydatid merely displaces the bronchi to one side and, although the lumena may eventually be narrowed, their lining ciliated epithelium is not destroyed and thus mucus, pus, and secretions do not stagnate; permanent inflammatory changes in the affected lobe are uncommon, at least until the parasite has been 'in situ' for a long time.

The growing cyst impinges upon the bronchi with which it comes into contact but even after a big parasite has been taken out of the lung it is unusual to find large bronchial fistulæ communicating with the empty space. As a rule there are one or two small fistulous communications, but these are lateral or side holes (*en bec de flûte*). In some cases the cyst ruptures into a bronchus and the membranes are coughed up, and in such the communication is larger.

The branches of the pulmonary arteries, which run with the bronchi, are also involved wherever the cyst comes into contact with them. It used to be thought that the episodes of hæmoptysis which occur so commonly in pulmonary hydatid disease were due to rupture of these vessels in the adventitia whose walls had been weakened by contiguity with the growing cyst. It was also thought that when a large cyst was removed by operation there was a risk of sudden hæmorrhage from some such vessel, left unsupported, in the adventitia. Serious hæmorrhage has not occurred in any of the operations in this series and yet the majority of the patients had had hæmoptyses at one time or another. Nor is it usual to find blood in the space which occurs after a partial rupture of the parasite and which lies between the hydatid and the adventitia. This space often fills with a crescent of air and is then known as a perivesicular pneumocyst; in its walls are the vessels concerned but we have not seen hæmorrhage into it. The most

probable explanation of the hæmorrhage which occurs in these cases is that there are abnormally large anastomoses between the bronchial and the pulmonary arterial radicals in the territory around the cyst and the bleeding is due to rupture of one of these minute subepithelial junctions. This explanation is accepted in cases of bronchiectasis, cystic disease, etc., and it is relevant to the surgeon because it means that massive hæmorrhage from the adventitia at operation is unlikely to occur.

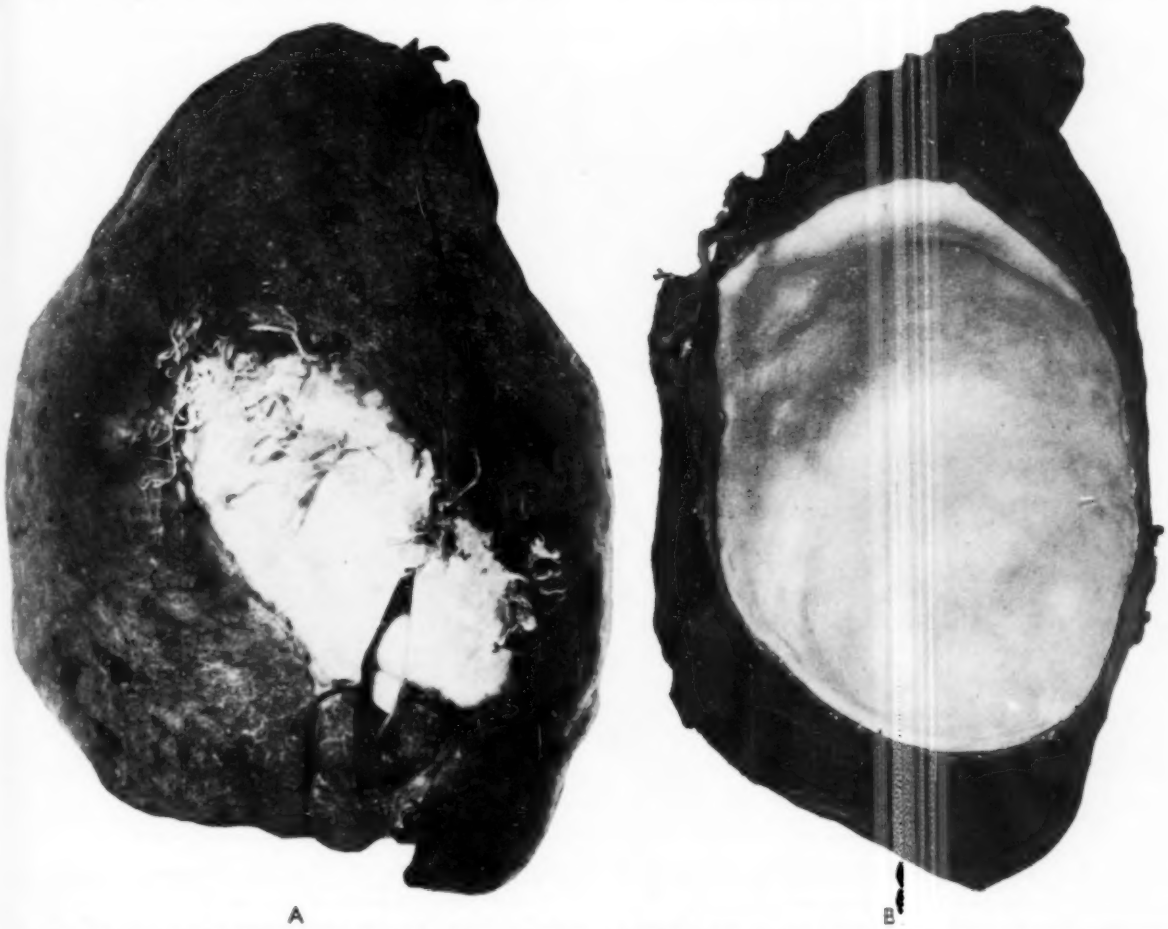


FIG. 282.—A, Large hydatid cyst in the left lower lobe of a woman, aged 55, who was suffering from bilateral pulmonary hydatid disease. The white area on the surface of the lobe is the pericyst or adventitia, and the laminated membrane of the cyst itself can be seen inside the latter. Note that, in spite of the size of the cyst, filmy pleural adhesions are only present in two isolated areas. B, The lower lobe has been cut open exposing the hydatid cyst. It occupies practically the whole of the lobe, and histological specimens showed inflammatory changes in the interstitial tissue and in the walls of the bronchi. (*Reprinted from "Thorax" Vol. 2, No. 1, March, 1947, Treatment of Pulmonary Hydatid Disease, by N. R. Barrett.*)

As the hydatid enlarges it eventually grows up to the visceral pleura. It may be that the point at which it surfaces is situated in one of the main fissures upon the mediastinal or diaphragmatic aspects of the lung. (*Fig.* 282.)

The first indication discernible to the naked eye is a prominence or bulge, the apex of which is greyish. This colour is due to the capsule of perivesicular atelectasis in the host and as the cyst continues to grow outwards it becomes less and less thick, so that in the end, it ceases to exist and the laminated membrane of the parasite is exposed, naked upon the surface. The laminated membrane is distinguishable from the adventitia because it is bluish or pearly white in colour and its surface is smooth, shiny, and moist. The hydatid is now free to herniate out of the lung into the pleural cavity.

The cyst, presenting upon the surface of the lung, evokes no inflammation in the adjacent pleura, unless inflammation has occurred from 'pneumonitis' in the lung around the cyst. A simple cyst is usually associated with a free pleural cavity and only filmy bands develop over the exposed adventitia. Moreover, if some form of two-stage operation is planned in which, as a preliminary, pleurodesis is assayed, the result may be disappointing because the exposed adventitia does not react to local irritants in the same way as the normal pleura, and adhesions between it and parietal pleura form sparsely.

It is often stated as a point of significance in the differential diagnosis that simple hydatid cyst does not cause rarefaction or distortion of the ribs or the vertebral column. We know of a cyst in the posterior mediastinum which 'eroded' the bodies of vertebræ, and in the present series simple hydatids were found to be 'eroding' the ribs on three occasions, and on this account were considered at first to be neurofibromata. The ribs over a cyst may be dented by the

'tumour' even though the pleural cavity between the two is free from adhesions.

It is difficult to explain why many of the patients suffer from localized pain, for it does not seem to be always due to pleurisy or to pressure upon bones. At operation there is no certainty that adhesions or other evidence of inflammation will be found, but upon the induction of a diagnostic artificial pneumoperitoneum. This test is of no value unless a good crescent of air can be demonstrated below the diaphragm, and sometimes the only way to be sure is to explore the 'tumour' surgically There is no place in the doubtful cases for diagnostic aspiration; this manœuvre not only may rupture the cyst and

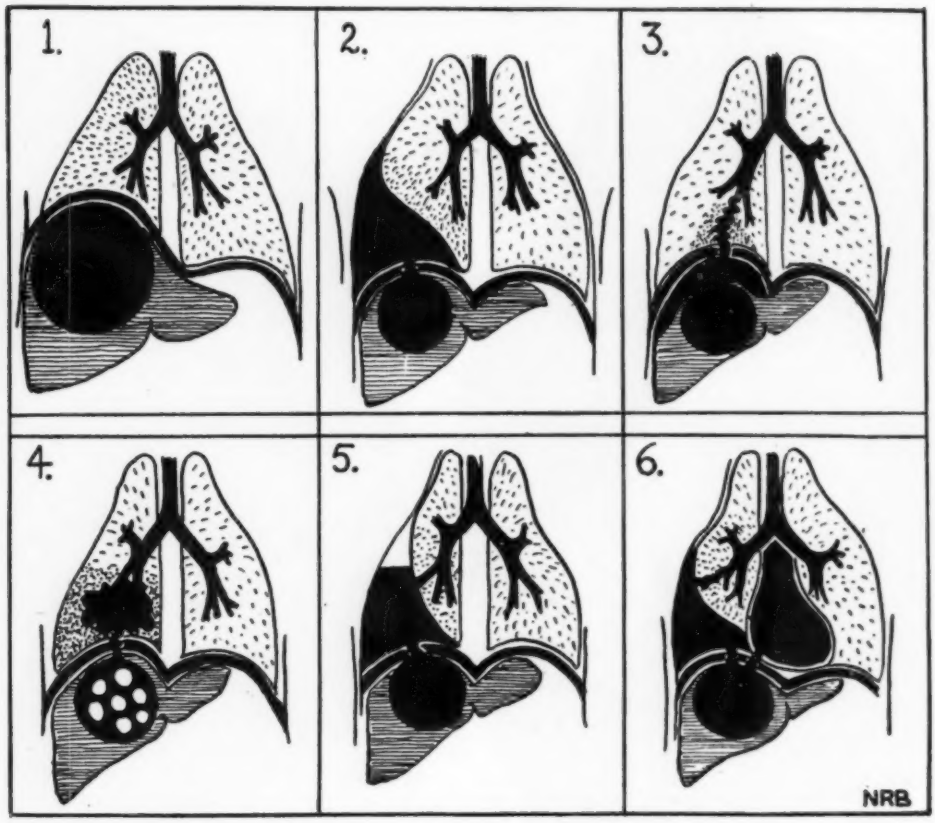


FIG. 283.—Diagrams to show the pulmonary and pleural complications of hepatic hydatid disease. 1, Elevation of diaphragm; 2, Empyema or 'water effusion'; 3, Subphrenic abscess and bronchial fistula; 4, Subphrenic abscess and lung abscess. Daughter cysts being coughed up; 5, Empyema and bronchopleural fistula; 6, Empyema and pericarditis or mediastinitis.

the pain is relieved by removing the hydatid from the lung.

Hydatid disease has not caused paralysis of the sympathetic, recurrent laryngeal, or phrenic nerves in any patient in this series, nor have there been any showing undoubted signs of vena caval or œsophageal obstruction. But there was one child, a girl aged 10, who for several weeks had a loud vascular murmur of the machinery type, which was most easily heard in the right paravertebral region just at the level of the hilum. This girl had a cyst in the right upper lobe which impinged upon the mediastinum.

Big hydatids do not always enlarge the hemithorax in which they are growing, but a bulging chest wall may occur in children. It has been said that a point of difference between a large basal lung cyst and another growing in the liver is that the former may cause a prominence of the affected side of the chest, whereas the latter displaces the diaphragm upwards. We have not observed this point of difference, and in distinguishing between cysts above and below the diaphragm have come to rely more disseminate the disease, but the patient may react to aspiration by anaphylactic shock.

It is a working rule that if multiple small daughter cysts are found in the lower part of the chest, they have generally originated in the liver. (*Fig.* 283.) The following brief history indicates the difficulty of diagnosis and treatment in this last type of case.

A woman, aged 32, was quite well until she developed a febrile illness and began to cough up "things like worms". She also complained of spasms of dyspnœa and palpitations which were put down to "heart attacks". She was eventually suspected, upon radiological evidence, of suffering from hydatid disease at the base of the right lung. The history of having lived on a Welsh farm and having kept dogs seemed to corroborate this opinion and the Casoni reaction was strongly positive. On admission her general condition was good, and there were no physical signs apart from those at the right base. The sputum contained no hydatid elements. Radiographs showed a lobulated opacity situated in the front of the right lower lobe and another similar shadow in the posterior basic segment on the same side. There was also a definite, calcified opacity in the top of the liver.

A pneumoperitoneum was induced but no air was seen beneath the right cupola of the diaphragm. (*Fig.* 284.)

The right lower chest was explored on the assumption that the patient had a dead hydatid in the liver and two secondary cysts in the lower lobe of the lung; but the pleural cavity and the subdiaphragmatic space was so obliterated by dense adhesions that it was difficult to be sure of any anatomical landmarks. A space about the size of a grapefruit was eventually opened. Its walls and large ones lobulated because they are distorted by any firm structure with which they come into contact. A characteristic of hydatid disease is the truly sharp demarcation between it and the surrounding normal alveoli. In hydatids as large and larger than an orange, the outline of the shadow seen in antero-posterior and lateral films is not circular. It is modified both by the proximity of adjacent structures,

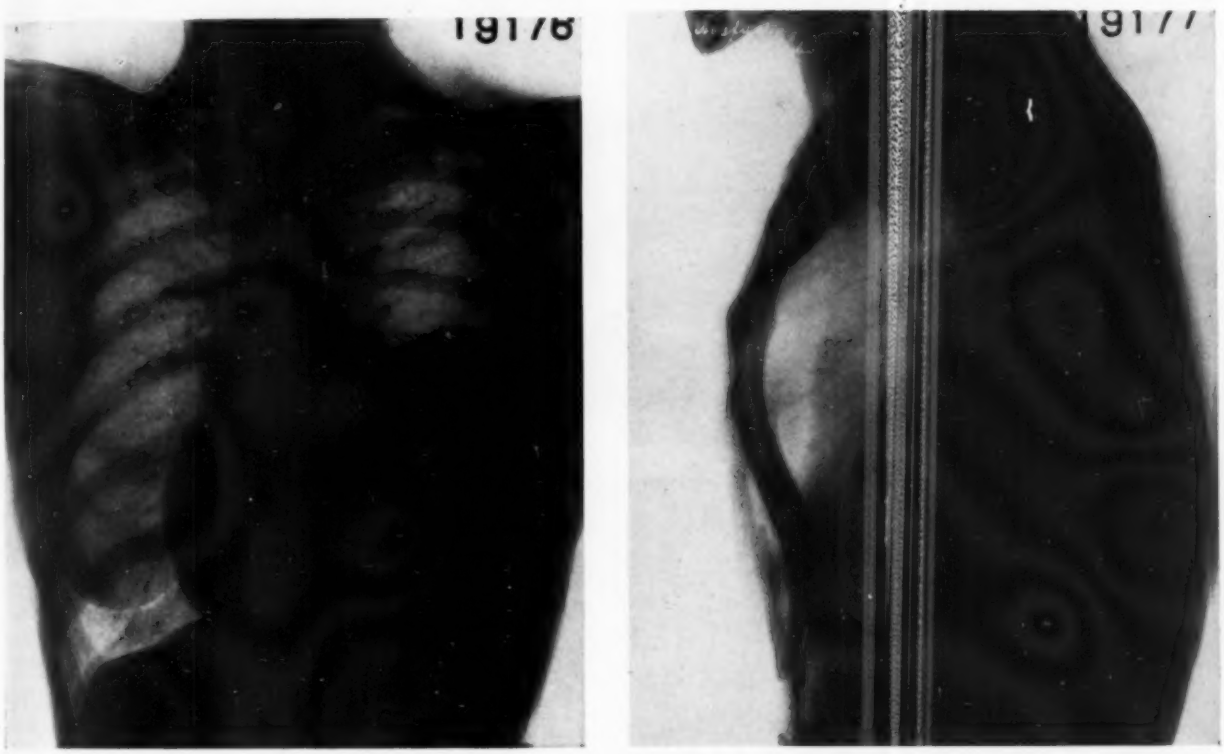


FIG. 284.—Anteroposterior and lateral radiographs showing the difficulty of knowing whether some hydatid cysts are above or below the diaphragm. This cyst lay above the diaphragm and the matter was settled by inducing a pneumoperitoneum.

were calcified and densely fibrous: it contained hundreds of daughter cysts which varied in size from a pea to a small orange: the cavity was not infected. It was evacuated as efficiently as possible but there were numerous ramifications. A pack was inserted and the wound continued to discharge hundreds of small daughter cysts for about 6 weeks. The discharge ceased and the wound healed. A second operation was done to remove the parasite at the back of the lower chest. Here again a large cavity was found. This contained much foul-smelling pus (cultures of which yielded *B. coli* and other intestinal organisms) and hundreds of small, and healthy, hydatid cysts. The cavity was evacuated and drained, and the patient made a rapid recovery to normal.

Six years later, after an intervening period of good health, she was again being investigated by her doctor but at this time there was no evidence of recurrence of hydatid disease although the Casoni reaction was still positive.

This patient undoubtedly suffered from a primary hepatic cyst which discharged through the diaphragm and and into the right lung.

There are many lesions in the lung which cause a circumscribed opacity upon radiology and hydatid disease is one of these. The shape of a hydatid cyst is not constant but varies according to its size and position. (*Fig.* 286.) Small cysts tend to be spherical, such as a large bronchi, and adapts itself to the chest wall or the diaphragm. If an artificial pneumothorax be induced in such cases the shape of the parasite in the collapsed lung may alter—not only as compared with the original films but according to the position in which the patient is examined. This point can be of diagnostic importance in assessing the likelihood of a circumscribed shadow in the lung being solid or cystic.

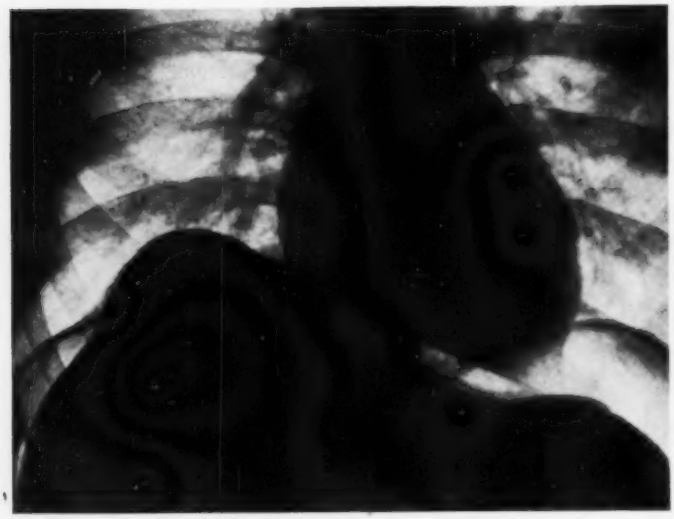
The Complications which can befall a 'Simple' Lung Cyst.—The most striking clinical feature of pulmonary hydatid disease is that unless, or until, some complication has overtaken the parasite the lesion is nearly always silent. The patient may experience transient symptoms of bronchial irritation, or vascular congestion (cough or mild hæmoptysis) but in many cases he comes to a surgeon because an unexplained circumscribed opacity has been detected upon radiography, in the lung: and it is in this particular group that exact diagnosis is often impossible before operation. It is also in this group that the Casoni reaction can be misleading in both positive and negative directions.

The principal factor which ultimately decides and regulates the fate of the cyst in the lung, and its final

effects upon its host, is the relation it bears to the bronchial tree.

In some cases the first intimation the patient has that he is harbouring a hydatid cyst in the lung is a sudden attack of symptoms due to anaphylaxis or to expectoration of peculiar sputum. In these cases the patient generally raises a considerable quantity of sputum which consists of mucus, frothy blood, hydatid liquid, and parts of the laminated membrane. The sputum is tenacious, it tastes salt, and causes violent paroxysms of cough. In other patients who harbour large parasites, the event of rupture passes off silently; even perhaps without symptoms, and the membranes are retained as a foreign body inside the adventitia. Between these two extremes all grades of clinical disturbance may be experienced, and, in a country like England where the disease is unusual, it may not be at all obvious—unless the diagnosis has been previously established—that the patient is trying to expectorate fragments of a hydatid cyst.

Sometimes when a cyst in the lung ruptures the patient dies suddenly, either during a violent spasm of coughing or for no apparent reason. In such a case part of the laminated membrane may be impacted



A

B

C

FIG. 285.—A, Anteroposterior radiograph showing a calcified hydatid cyst in the liver of a man. A pneumoperitoneum has been induced which proves that the lesion is below the diaphragm. B, The same patient as in A. Lateral radiograph. This shows another small cyst lying anteriorly. C, Another patient showing that a very penetrating radiograph may reveal a calcified hydatid in the liver.

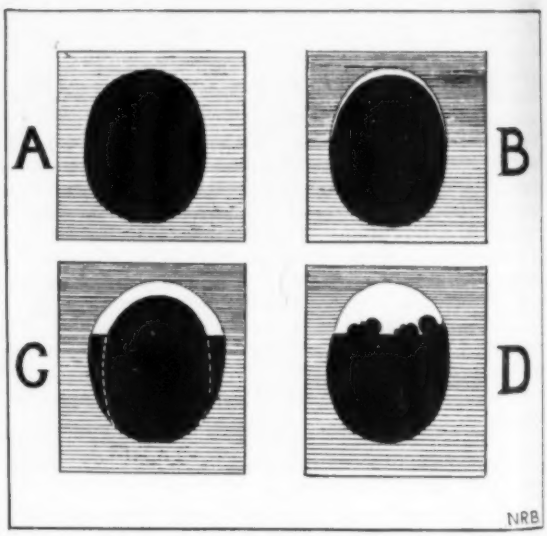


FIG. 286.—Diagram showing the four principal radiological appearances which occur in pulmonary hydatid cyst. A, Simple cyst; B, Perivesicular pneumocyst; C, Ruptured cyst; D, Hydatid abscess with the ruptured membranes floating in the liquid.

in an obstructed larynx, or death may have resulted from sudden vagal stimulation causing cardiac asystole. The latter phenomenon is not peculiar to hydatid disease, but is associated in some way with the irritation which any small object, impingeing upon the larynx, may cause.

We have not seen serious symptoms due to anaphylaxis except in one patient. This was a soldier who had two hydatids in the lungs. The diagnosis was not made at the time and the doctor aspirated one of the opacities to see what it contained. He drew off fluid like spring water and guessed that the lesions might be hydatids: he then proceeded to test the matter out immediately by injecting a drop of the fluid into the dermis of one arm! Within a few minutes the whole arm and side of the chest and neck had swollen up, the patient was profoundly shocked, and had rapid respirations and tachycardia. It was some time before equilibrium was re-established.

One might think that the rupture of a lung cyst would generally be the result of some crushing force transmitted through the chest wall to the lung. This is not so. The event is rarely associated with trauma: it happens spontaneously. Nor is there any method of forecasting which cysts will rupture and which will grow to giant size. Small cysts are not likely

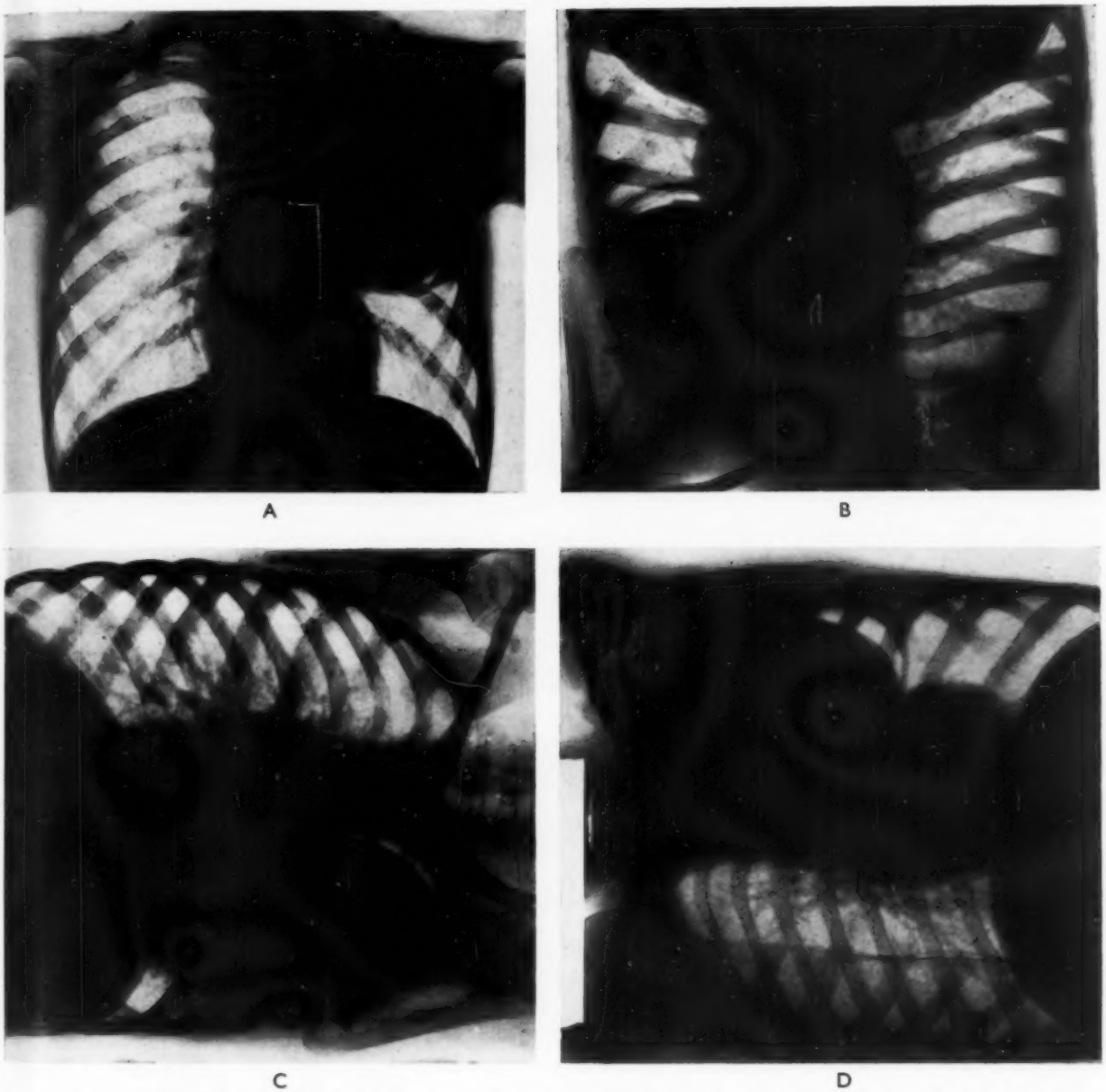


FIG. 287.—These four radiographs are all of the same young patient, who came up to hospital and was found to have a hydatid cyst in the left upper lobe of the lung—they show that the crescent of air in a perivesicular pneumocyst shifts to the top of the opacity whatever the position of the patient may be. A, The patient is erect and the air is just below the apex of the lung. B, The patient has been radiographed in the head-down position and the crescent of air is now between the opacity and the diaphragm. C, The patient is lying on the left side and the air is against the mediastinum. D, The patient is lying on the right side and the air is against the chest wall.

to rupture although one did in this series ; it occurred in a child of 6 and was no bigger than a plum at the time. If a cyst in the lung ruptures and its contents are partially evacuated by way of the bronchial tree the patient may then enter a period of symptomatic quiescence, in which he and his physician believe that a cure has been achieved. This latent period exists in such a case because the disintegrating

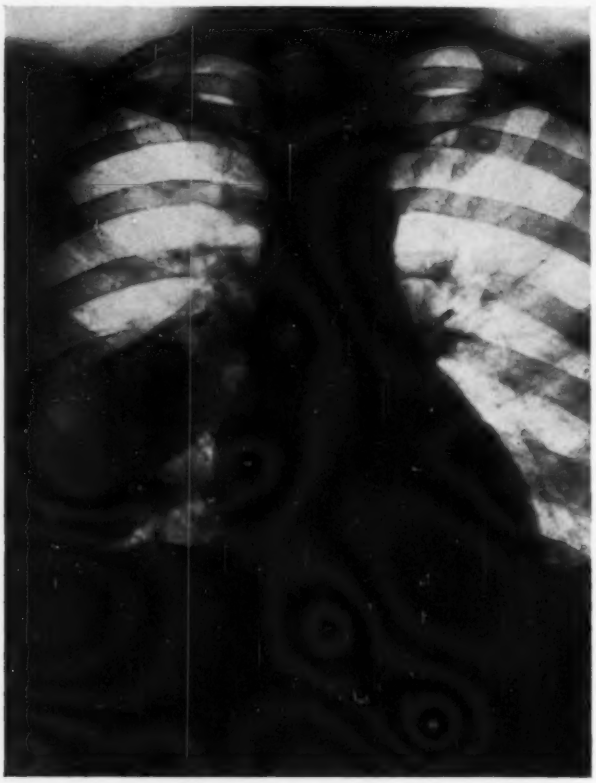


FIG. 288.—This radiograph shows a cluster of daughter cysts in the right lower lobe. A year previously the patient had had 2 simple cysts removed, intact, from the same cavity in the right lower lobe. The daughter cysts began to grow about three months after the first operation. The treatment in the end was lobectomy and the patient has remained well.

membranes do not cause symptoms until infection of the cavity and its retained foreign bodies supervenes.

Rupture can be partial or complete.

A partial rupture is one in which a little liquid has escaped through a small tear in the laminated membrane into the space between it and the adventitia, but the life of the parasite is not seriously threatened. Whenever this happens the laminated membrane begins to separate from the adventitia and the space so formed fills with a little fluid from the cyst. Very often the local retraction of the laminated membrane from the adventitia lays bare a small bronchial fistula in the adventitia and air enters this space ; it can be detected in radiographs as a crescent like a new moon. This crescent, which is practically diagnostic of hydatid disease, is called a 'perivesicular pneumocyst'. It indicates quite certainly that a partial rupture has occurred and it can be distinguished from other lesions, such as breaking down peripheral tumours which can show a somewhat similar appearance, because the crescent of air is always at the top of the shadow irrespective of the position in which the patient may be photographed. This point is well shown in *Fig.* 287.

In most of these cases the small perforation in the laminated membrane seals off spontaneously and the life of the parasite proceeds as before. These patients may have consulted a doctor for an irritating cough or for some transient allergic manifestations, but in many the accident to the parasite passes unnoticed. They do not cough up enough liquid to remark upon it. After such an episode the parasite can remain as a single univesicular hydatid ; but if active hydatid elements have escaped into the perivesicular pneumocyst, then a second, or perhaps a third, hydatid may grow within the original adventitia so that the surgeon may find simple daughter cysts when an operation is first performed. Such daughter cysts are limited in number and do not grow in hundreds as they do in the pleural cavity or the liver. In pulmonary hydatid disease it is uncommon to find multiple simple cysts within a single, uninfected, adventitia. (*Fig.* 288.)

A complete rupture threatens the life of the parasite and may prove fatal to the patient. The least severe cases are those in which the laminated membrane tears little by little and the liquid is discharged slowly into the bronchial tree. Whenever this happens two things are certain. The first that the laminated membrane has turned inside out, and the second that its contents have been discharged into the bronchial tree. Rupture of the hydatid practically always opens up a bronchial fistula between the 'sac' and the respiratory passages, and the lesion now shows on radiographs as a cystic space in the lung containing air, with, perhaps, a little fluid still left in the cavity. Most of the fluid will have been expectorated. In some cases parts of the curled up and torn laminated membrane can be seen floating in the fluid, and this appearance has been likened to that of a water-lily floating just above the surface of a pond. It is called the 'water-lily' sign but is not pathognomonic of hydatid disease.

We had occasion recently to observe a patient in whom a simple cyst ruptured into the adjacent bronchus ; but instead of the hydatid collapsing, as described above, the laminated membrane itself became distended with air and presented radiologically as a lung cyst. (*Fig.* 289.) At operation a hydatid membrane, apparently intact and filled tensely with air, was removed. This could only have been a temporary state because the parasite could not live unless the air was eliminated and replaced by hydatid fluid.

The way in which rupture occurs importantly influences the clinical symptoms which afflict the patient. If a large cyst suddenly bursts, the liquid is discharged into the adjacent bronchi ; in this event bronchial or tracheal obstruction may result and be fatal. More usually the patient becomes acutely ill with the manifestations of partial bronchial obstruction to which may be added those of anaphylaxis. It is common for fragments of the membranes to be coughed up. These are generally described by the patient as being " pieces of matter " or " flesh " and are not the classical 'grape skins'. 'Grape skins' are small but complete or ruptured daughter cysts, and since multiple small daughter cysts are unusual

in the lung they are rarely seen in the sputum. The tissue expectorated is disintegrating fragments of the laminated membrane. These fragments, if small, can be disposed of by coughing, but if curled up into plugs of tissue, they impact. In one case in this series Sir Clement Price Thomas removed such a

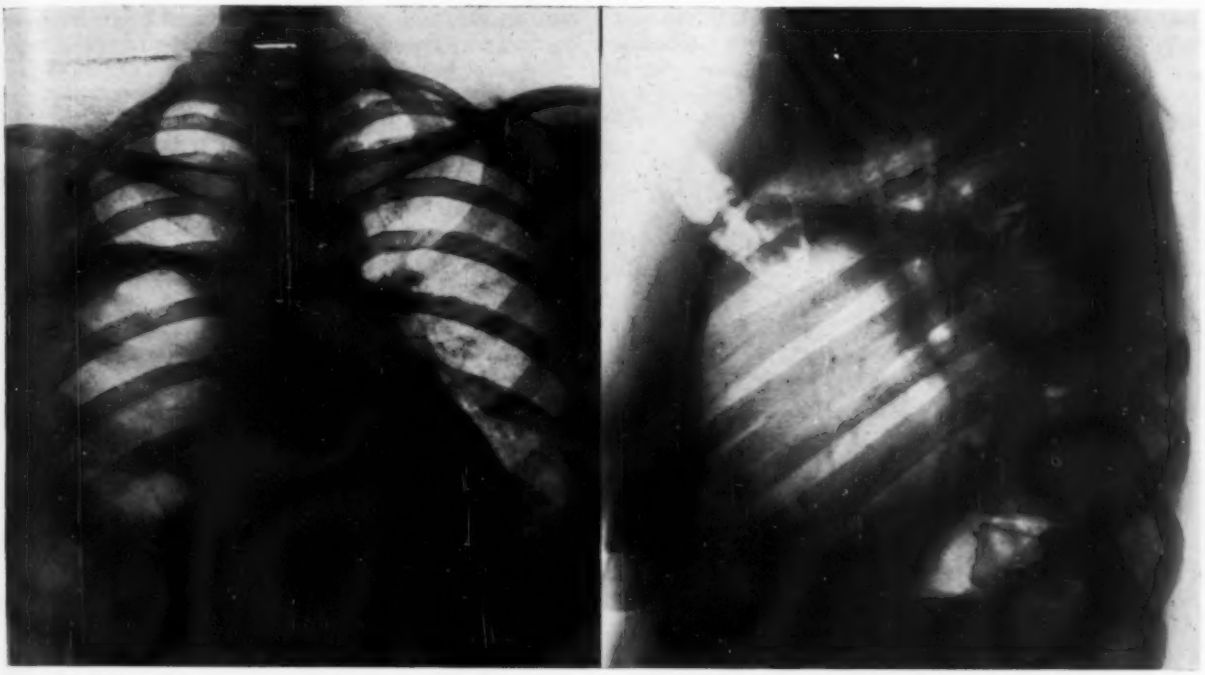


FIG. 289.—These are the radiographs of a woman, aged 32, who was admitted to Morriston Hospital, April, 1951, with a 4 weeks' history of cough with mucopurulent sputum, and 3 weeks right pleuritic pain. The patient was in fairly good general condition. The Casoni test was strongly positive. Chest films show 2 air-containing cavities in the right lung, the larger one showing a perivesicular pneumocyst. At right thoracotomy (D. M. E. Thomas) the larger cyst was removed intact from the right lower lobe, and was found to be completely air-filled. Later, on puncturing the cyst it slowly deflated. The smaller infected cyst was also removed. Post-operative course uneventful. Presumably the larger cyst had originally ruptured into a bronchus, emptying itself of fluid, and later the tear in the laminated membrane had become sealed off again.

FIG. 290.—This photograph has been made from a section of the lower lobe of a patient suffering from a hydatid abscess. Note the curled-up and disintegrating laminated membrane of the original hydatid cyst.

foreign body by bronchoscopy, from the lower-lobe bronchus :

> . . . it had lain in this place for some months. The patient was a man, aged 37, who first noticed cough and pain in the chest in November. A diagnosis of ' pneumonia ' was made, and the illness did not resolve. He developed persistent cough, sputum, and hæmoptysis, and was eventually X-rayed, and found to have a circumscribed opacity in the right lower lobe. At bronchoscopy, which was done 6 months after the onset of symptoms, some coiled-up fragments of the laminated membrane were impacted in the lower-lobe bronchus and were drawn out, relieving pus, bottled up behind the obstruction. The affected lobe was removed later, and the remainder of the membranes of a ruptured cyst were found lying loosely in a septic lung cavity.

If the parasitic membranes have been coughed out of the lung completely the remaining space, which is called the " empty sac " generally obliterates rapidly ; but if fragments of membrane remain they act as dead sequestra or as foci from which daughter cysts grow. The essential difference between hydatid cysts in the lung and those occurring in abdominal viscera is that the former are in communication with the bronchial tree from the moment rupture occurs, and thus are particularly liable to get infected.

When parts of a dead hydatid are retained the patient develops a chronic lung abscess around the foreign body and if this is allowed to proceed the

inflammation spreads to engulf the whole lobe, and ultimately develops into a spreading pneumonitis. This sequence was well shown by a patient who was known to harbour a simple univesicular cyst; he elected to ignore the parasite as it was not causing symptoms at the time, but within 18 months the cyst had ruptured and been partially expectorated. When ultimately treated by lobectomy the lower lobe consisted of a small, solid, mass of inflammatory tissue containing the remnants of the dead hydatid cyst inside an abscess. The lobe had been destroyed beyond any possibility of natural repair. (*Fig.* 290.)

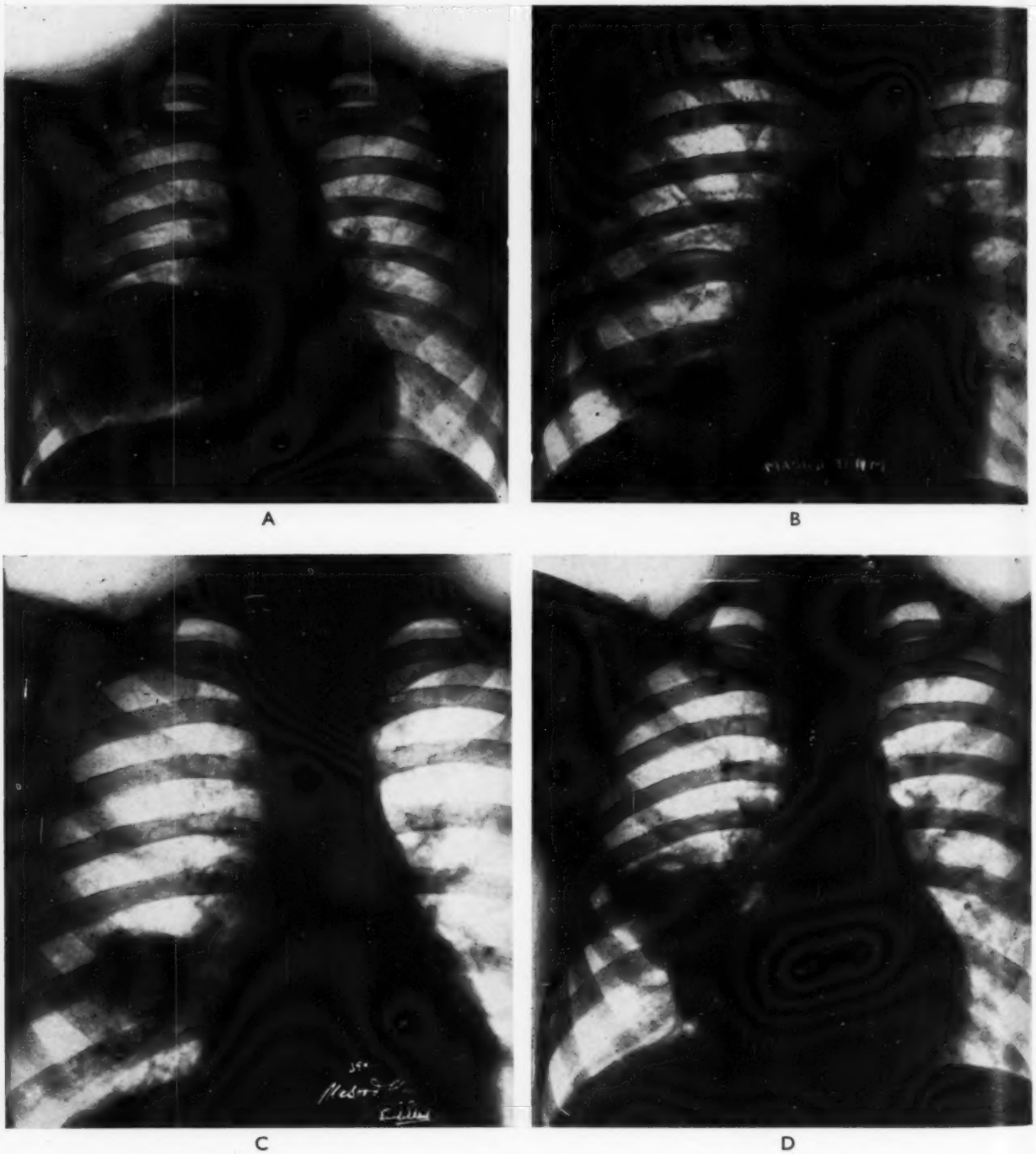


FIG. 291.—These four films show the development of hydatid disease in a man, aged 19, who came from Pakistan. A, There is a solitary 'simple' hydatid cyst in the right lower lobe of the lung. The patient was symptomless, except for a mild cough, at the time. B, Thirty months later a radiograph shows that the parasite has ruptured. In this film there is a fluid level inside the adventitia (the original capsule in the host) and, breaking the surface of this fluid level, are the remnants of the curled-up laminated membrane (the 'water lily' sign). The patient had blood-stained sputum. C, Nine months later the patient is still coughing up blood-stained pus, and the adventitia has contracted down so that, at first sight, the patient seems to be getting better—this is not the case, because some hydatid elements have, in fact, survived and daughter cysts are forming. D, One year later the patient has a growing 'hydatid abscess' and this was proved at operation to contain multiple daughter cysts.

Some hydatid fragments, capable of reproducing hydatid disease, often remain alive for a long time after serious complications have irrevocably destroyed the parent cyst. These fragments of the germinal membrane and brood capsules may give rise to new hydatid cysts even if the sac in which they are contained has become infected. In such a patient the lesion will at first diminish in size after the simple cyst has ruptured. (*Fig.* 291.) This is due to evacuation of the fluid it contained. It will then present the clinical and radiological features of a subacute or chronic lung abscess and, as the daughter cysts begin to grow, the opacity in the lung will enlarge again. In one of our patients a simple hydatid cyst was removed from the left upper lobe. At operation some of the hydatid fluid was inadvertently spilt into the lung cavity from which the parasite had been taken out. In spite of this accident it seemed at first as though the patient had been cured, but three months later the opacity in his upper lobe was still present and he was coughing up pus. One year later the lesion was increasing in size and when the lobe was eventually removed it was found to contain a chronic lung abscess in which there were dead fragments of the original laminated membrane and several live daughter cysts. He has been cured by segmental lobectomy.

Two patients in this series first came to hospital with the signs and symptoms of acute empyema. The empyemata were drained and several days later the first of hundreds of small hydatid cysts began to be discharged through the drainage-tubes. In both cases a cyst had ruptured into the pleural cavity and secondary infection had occurred. In one the primary hydatid cyst was situated in the lung and in the other it was ultimately found to be in the liver. This condition is called ' hydatid empyema '.

Some radiologists have stated that calcification does not occur in the adventitia around a hydatid cyst. This is not true. Calcification has been found on several occasions in the present series. (*Fig.* 292.) It occurred in the adventitia of complicated cysts and looked like the thin shell of an egg. When calcification is found this does not indicate that the hydatid is dead. It may be, but it may not. In viscera such as the spleen it is common to find radiological evidence of calcification and in such the finding generally means that the parasite has died and can be ignored. In the lung the calcified hydatid is almost certainly in communication with the bronchial tree somewhere, and thus is probably infected and dangerous.

FIG. 292.—Radiographs of a woman showing marked calcification in the adventitia of a hydatid cyst. This patient has a hydatid abscess in the lung and was successfully treated by lobectomy. (*See also Fig.* 279, in which there is a calcified hydatid in the lung of a child.)

STATISTICS CONCERNING THE PATIENTS IN THIS SERIES

The present series comprises 71 patients who had at least 94 cysts in their lungs. Forty of these were males and 31 females. About 10 per cent had contracted their disease outside the British Isles.

From the histories of those patients who contracted hydatid disease in the British Isles certain points may be deduced. There are districts, and even villages, in which the risks of infestation are high, whereas throughout most of the country there is no risk. It cannot be said that all cases have occurred in backward or isolated rural areas, for a number of patients have not lived in the country and presumably acquired the disease from dogs in towns. In this connexion the present popularity of greyhound racing and whippet coursing may be relevant ; and in some areas the regulation of abbatoirs is so lax that

scavenger dogs can easily get access to infected offal. About 10 per cent of the total number of patients gave no history of having been especially connected with dogs and about 25 per cent had not lived in the country.

The ages of the patients at the time of their admissions to hospitals are given in *Fig.* 293.

The oldest patient to be affected was a man, aged 62, who had a single simple cyst which was successfully treated by lobectomy; the youngest was a boy,

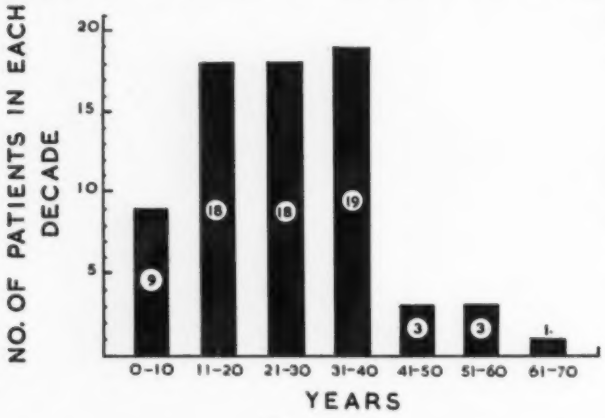


FIG. 293.

aged 3 years and 11 months, who was cured by removing a simple cyst intact.

The Anatomy of the Cysts in the Lung.—Fifty-two patients had a solitary cyst in the lung. The remaining 19 patients were found to have more than one cyst in the lung. Amongst the latter there were 6 patients in whom there were two cysts in one lung —the other lung being clear—and 13 patients who harboured bilateral cysts. There were 4 patients who were found to have three simple lung cysts at the same time, but none in whom this number was exceeded.

Amongst the 9 children who were less than 10 years old, there were 4 who had more than one cyst in the lungs. One child, a little girl aged 6, harboured two parasites, one of which was simple but the other had ruptured and was already infected.

In 5 cases the parasite had herniated out of the lung and given rise to secondary echinococcosis in the pleural cavity: and in another 8 there were cysts in the liver as well as in the lungs. The spleen was only involved on 2 occasions as far as we could tell. One patient had had hydatids in the muscles of the chest wall after marsupialization and drainage of a lung cyst in New Zealand, and there was a history of hydatid cyst having been removed from the sternomastoid muscle. One patient developed multiple hydatids in the peritoneal cavity after successful removal of a parasite from the lung.

The relative frequency of hydatid disease developing in the lung as compared to the liver is a matter of interest. Our experience does not give the answer to this problem because most cases referred to us have thoracic diseases. Nevertheless we feel that it cannot be true that liver hydatid disease is ten times more common than lung cysts, for this would indicate that during the years when our cases were treated a great many abdominal cases should have been treated in the British Isles.

The records of St. Thomas's Hospital show that, since 1937, the following cases have been treated in the hospital.

Echinococcus of lung		4
"	" pleura	1
"	" kidney	2
"	" oviduct	1
"	" liver	4
	Total	12

and the Registrar-General's Statistics give the number of patients dead from hydatid disease in England and Wales between the years 1940 and 1949 as:—

	Liver		*Other or unspecified organs*	
	Male	Female	Male	Female
1940	8	7	6	7
1941	3	6	9	3
1942	2	8	6	1
1943	5	1	3	1
1944	2	6	5	1
1945	9	6	2	3
1946	4	6	3	1
1947	5	7	4	1
1948	6	4	2	7
1949	5	5	1	4
	49	56	41	29
Total Liver	105		Total Other	70

At the time we first saw our patients the pathological state of their lesions was as follows. In 48 the hydatids were 'simple'; in 23 they were 'complicated'. In a further 12 we found 'simple' and 'complicated' cysts present in the lungs of the same patients; and in 8 there were daughter cysts, some of which were the result of imperfect operations and some had occurred spontaneously.

Length of History.—This is shown in *Fig.* 294.

From these statistics we conclude that hydatid disease, and particularly lung cysts, are not uncommon in the British Isles. Men have been

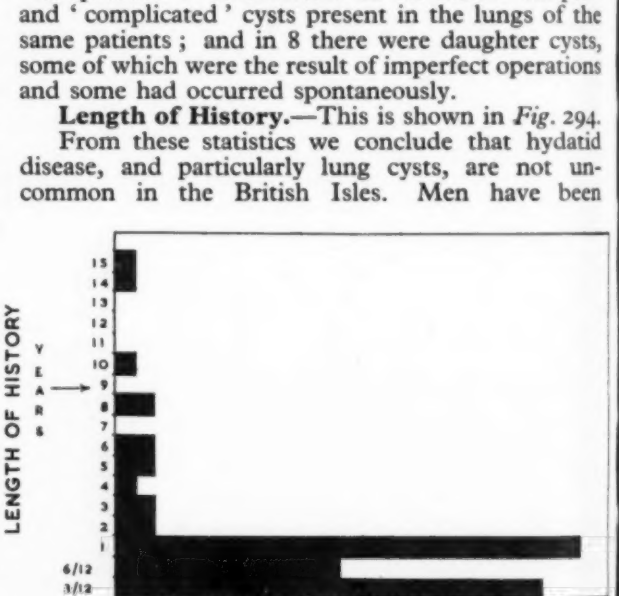


FIG. 294.

affected in about the same proportion as women, and children under the age of 10 were certainly not immune. On the average the clinical histories of our patients have been short, and in two-thirds of the cases the cysts were 'simple' when the patients were first treated. The majority were suffering only from pulmonary hydatid disease but in 8 instances the liver was also involved. In our experience systemic infestation has been limited to 4 patients.

THE TREATMENT OF PULMONARY HYDATID DISEASE

A hydatid cyst, living in the lung of a human being, can only produce one of two end-results. Either the cyst must die or be removed surgically, or the patient must ultimately suffer from serious and, possibly, fatal disease. A dead, retained, cyst is unlikely to prove an innocuous foreign body. The aim of treatment must therefore be—removal of every cyst as soon as the diagnosis has been made.

Nothing which has been written in the last few years convincingly suggests that lung cysts should be treated medically or conservatively. Some have tried to avoid operation by aspirating simple cysts, after inducing pleural adhesions, and injecting acid pepsin into the cavity containing the ruptured membranes. The rationale is said to be that the enzymes fragment the laminated membrane so that it can be coughed up. This method seems to be hazardous, uncertain, and to be based upon an incorrect assumption, namely that acid pepsin will digest the laminated membrane.

The real and fundamental objection to medical or expectant treatment is that there is no certain way of getting *all* the disintegrating laminated membrane out of the cavity of the lung, and no possible way of assessing the completeness or otherwise of spontaneous expectoration—except by watching events and studying serial radiographs. Now although a simple cyst may be relatively harmless to the lung, a suppurating cyst is certain ultimately to set in motion the whole train of destructive pathological reactions which can be induced by a chronic lung abscess. These changes cannot be influenced by antibiotics or other drugs because there is a foreign body retained in the cavity. If a patient expectorates a cyst, and is cured, it can be assumed that the membranes have been *totally* voided.

In the management of simple hydatid disease three things must be achieved : the parasite must be removed, the wound and adjacent pleural cavity must not be contaminated with live hydatid elements, and some provision should be made to deal with the 'empty sac' which remains in the lung.

Simple hydatid cysts should be removed from the lungs as soon as they are diagnosed. In the past this was not always considered to be practicable, because it was felt that it was not safe to incise through the lung to reach a deeply placed hydatid. Recent experiences with operations such as segmental lobectomy, performed for other lesions, have shown that by keeping to the intersegmental planes it is easy to expose 'tumours' in the lung, however deeply they may be placed. Thus there is no longer any need to defer operation on the grounds that the hydatid is situated deep in the lung.

Nor should operation be deferred in the remote hope that the whole parasite may be expelled by cough; for, at best, this event is uncertain and dangerous. As far as we can ascertain, nobody has, as yet, produced figures to show all the dangers of awaiting spontaneous rupture; but they are certainly greater than those of thoracotomy by modern methods. Moreover, expectant treatment may fail or may leave the patient suffering from progressive pulmonary suppuration, whereas a cure can be almost guaranteed by surgery. The fact that a patient happens to harbour multiple parasites in the lungs is an added reason for early surgical treatment.

Up till recently the argument concerning the precise details of how to get the simple parasite out of the lung has ranged about the relative merits and safeties of a two-stage operation (in which the lung in the vicinity of the parasite was marsupialized to the chest wall) and secondly, of removing the hydatid, after puncture or aspiration, across the open pleural cavity. We submit that both operations are bad and should now be given up. They were adopted at a time when the hazards of thoracotomy, and particularly those of the possible post-operative complications, were paramount. These hazards included tension pneumothorax, acute pyopneumothorax, acute anaphylaxis, and chronic empyema. They glossed over the inevitable fact that whether you puncture the cyst, or aspirate the cyst, the risk of contaminating the exposed tissues is great. It is assumed nowadays that to spill a large, or a tiny, amount of hydatid fluid is absolutely undesirable.

Another operation which we have advocated and used in the management of simple cysts was lobectomy. This is now seldom used even in the management of large hydatids. In the past we argued that large hydatid cysts, being space-occupying lesions in a lobe, must inevitably destroy that lobe by pressure upon, and distortion of, adjacent bronchi. We now realize that even large hydatids may do remarkably little permanent damage to the lung and that simple removal practically always results in restitution to normal in a remarkably short time. This point has been emphasized by Australian colleagues from the first, and it was they who urged us to restrict lobectomy to those cases of simple hydatid disease in which, at thoracotomy, it was quite obvious that permanent and irretrievable changes had occurred in the affected lobe. In practically all patients suffering from simple pulmonary hydatid disease it suffices to remove the parasite.

How to get the Parasite out Intact.—The ideal operation is one which succeeds in removing the parasite from the lung intact. In this way all possibility of contaminating the wound, the pleural cavity, or the adventitia in the lung are avoided.

There are a variety of points which bear upon this problem. Any operation in which the membranes are deliberately punctured and a "strong sucker plunged into the cavity to remove the liquid and the hydatid" must result in contamination of the exposed tissues. It has been shown that in practically all cases the hydatid fluid from a simple human cyst contains live elements. Similarly operations in which the parasite is first 'aspirated dry' with a fine needle generally end up with rupture, and once again the wound and the surgeon's hands are contaminated.

Nevertheless procedures of this type are in general use to-day and the results claimed, both immediate and late, are said to be good. These techniques have been used sometimes in the present series, and, in retrospect, it has been surprising to us that more of the patients did not develop secondary echinococcosis. But 6 patients out of less than 50 treated in this way did get daughter cysts and for this reason we have given up removing the parasite by any technique which employs aspiration or rupture.

To meet these demands one of us (N. R. B.) devised an operation which imitates the natural sequence of events when a parasite herniates out of the lung into the pleural cavity. The operation is applicable to single cysts or to multiple cysts, and on several occasions has been applied bilaterally. It has no place in the management of ' complicated ' cysts.

The patient is given a general anæsthetic, and this is maintained after a cuff tube has been introduced into the trachea to make an air-tight circuit. The head of the table is lowered so that if the cyst ruptures into the bronchus the liquid drains from the lungs by gravity. The appropriate hemithorax is opened, either by rib-resection or an intercostal incision placed at about the level of the middle of the cyst, and the pleural cavity is opened *widely* with rib-spreaders. If pleural adhesions are present they are divided so that the lobe which contains the cyst is free. The affected lobe, or, where a giant cyst is present, the cyst, is then enclosed in a bag of macintosh, made like a sponge bag, with strings at the top for gathering up the inlet, but the bottom is cut open. The bag is placed round the lobe containing the cyst with the strings at the hilum, and these are drawn up to encircle the neck of the lobe without impeding respiration in the lobe or the blood-supply. The bottom of the bag is then opened out, brought through the incision and spread out on the surface of the wound so that the affected part is inside a waterproof container. The bag not only gives some protection against accidental rupture but, later in the operation, is an excellent retractor. If such a bag is not available an ordinary piece of macintosh tucked in behind the cyst will serve. The exact position of the cyst in the lobe is now ascertained and in most cases the adventitia will be seen on the surface of the lung as a patch of white membrane. If the hydatid has not grown up to the surface, the lung is incised along one of the inter-segmental planes over the cyst and down to the adventitia. The stage is now set for removing the cyst.

The adventitia is partially incised by a straight cut with a knife. The length of the incision should be about the same as the diameter of the cyst measured in the radiographs, and it should be placed towards the bottom of the cyst as the patient lies on the table. The incision must be made cautiously, because as a rule the adventitia of lung cysts is not more than $\frac{1}{8}$ in. thick. The object is to form a crack in the adventitia and not to expose the laminated membrane throughout the length of the incision. When the laminated membrane is exposed at any point the fact will be obvious because of its colour—white, faintly tinged with blue. Once the laminated membrane has been exposed in one spot the incision in the adventitia can be extended by lifting it off the cyst and cutting the lung in several directions with blunt-pointed scissors. If the method has been used correctly the cyst begins to herniate out of the lung.

The surgeon's job is now for the time being over; success depends on not touching the parts until the cyst has been extruded from the lung. In particular neither the laminated membrane nor the adventitia should be touched or grasped with toothed forceps. Extrusion is achieved by the anæsthetist who steadily increases the pressure in the anæsthetic circuit so that little by little, the hydatid is gradually extruded from the lung intact, and lies in the macintosh bag. This happens for two reasons. The first is that, as the anæsthetist inflates the lung, the slender tissues over the cyst are stretched and torn, thereby exposing a greater and greater area of the dome of the cyst; and secondly, some gas escapes from tiny bronchi in the adventitia and gets into the space between the parasite and the host, thus providing an additional *vis a tergo*. During this period there must be no attempt to hurry matters, and if the surgeon cannot be persuaded to leave well alone the safest instrument for him to use is a sterile household spoon of appropriate size.

Once the cyst has herniated out of the lung it lies in the bag by the side of the lobe which originally contained it. The operation table should now be tilted towards the surgeon so that by tightening the lower margin of the bag and by gently introducing the hand behind the extruded parasite, it can be lifted from below so that it rolls naturally into a porringer held to receive it. The moment when a hydatid rolls out of the lung intact is one of the most satisfying in surgery.

The method described above is suited to the treatment of hydatid cysts which are not too large; but it has been our experience that the chances of getting out a parasite the size of a grapefruit, for instance, in this way are not good. Too often the laminated membrane, which is old and stretched, begins to split under the strains imposed by the weight and bulging of the liquid it contains, and once this starts the hydatid is bound to rupture. Moreover, the manipulations necessary to juggle a large parasite out of the lung demand more lifting and pushing than in the case of young cysts which simply drop out.

The best way to get a large cyst out intact is to remove it in the plane between the adventitia and the surrounding normal lung. That is, take out the parasite together with the capsule formed by atelectatic lung and fibrous tissue which surrounds it. In practice this is not difficult because the capsule formed from the lung is itself sharply demarcated—even in large cysts—from the surrounding alveoli and a plane of cleavage, which is seldom vascular, can readily be found. The larger the cyst the easier it is to enucleate it with its adventitia intact.

With patience and some manual dexterity practically all ' simple ' cysts—whatever their size—can be removed intact and without risk of further infestation. But, sometimes, whilst enucleating a parasite in one of these ways it ruptures unexpectedly and, to meet this possibility, the surgeon should have a strong suction apparatus available and in working order.

In no case in this series has serious hæmorrhage from the wall of the cavity been encountered at the moment of enucleation of the parasite. This complication, if it were to occur, should present no difficulty if the surgeon has the chest wide open, for he need only grasp the hilum of the affected lobe, and having thus compressed the vessels and staunched the flow, he can pick up the bleeding point at leisure.

The ' Empty Sac '.—After a hydatid has been enucleated from the adventitia the next problem is what to do about the residual space. It has already been stated that the membrane which forms the wall of the cavity is composed of compressed, atelectatic, alveoli if the cyst has been small, and fibrous tissue if it was large. After enucleation of a hydatid one or more bronchial fistulæ can generally be seen opening ' *en bec de flûte* ' upon the walls of this space. If the surgeon takes no steps to close these off the patient develops a tension pneumothorax and probably an empyema. To deal efficiently with such fistulæ—and in our experience one or more is always present—depends upon the knowledge that pulmonary hydatid disease is primarily a segmental lesion. The bronchi against which any single hydatid impinges are probably all branches of one main segmental bronchus. This main bronchus is situated near to

that part of the cyst which approaches most closely to the hilum, and sometimes it is itself affected and open.

If, after enucleation of the parasite and dealing with the fistulæ, the empty space is simply closed by sewing up the incision through the adventitia, one of several things can happen. In some cases the dead space remains as a tension cavity, in others it fills with exudate or blood; but in the majority, and particularly in children, it will disappear spontaneously in a few weeks.

A feature of these residual spaces is that they can remain empty for months even though distended with air. In one patient, a little girl who harboured bilateral, large, simple cysts, the empty space obliterated slowly over a period of 18 months on the right side, but had disappeared completely in 3 months on the

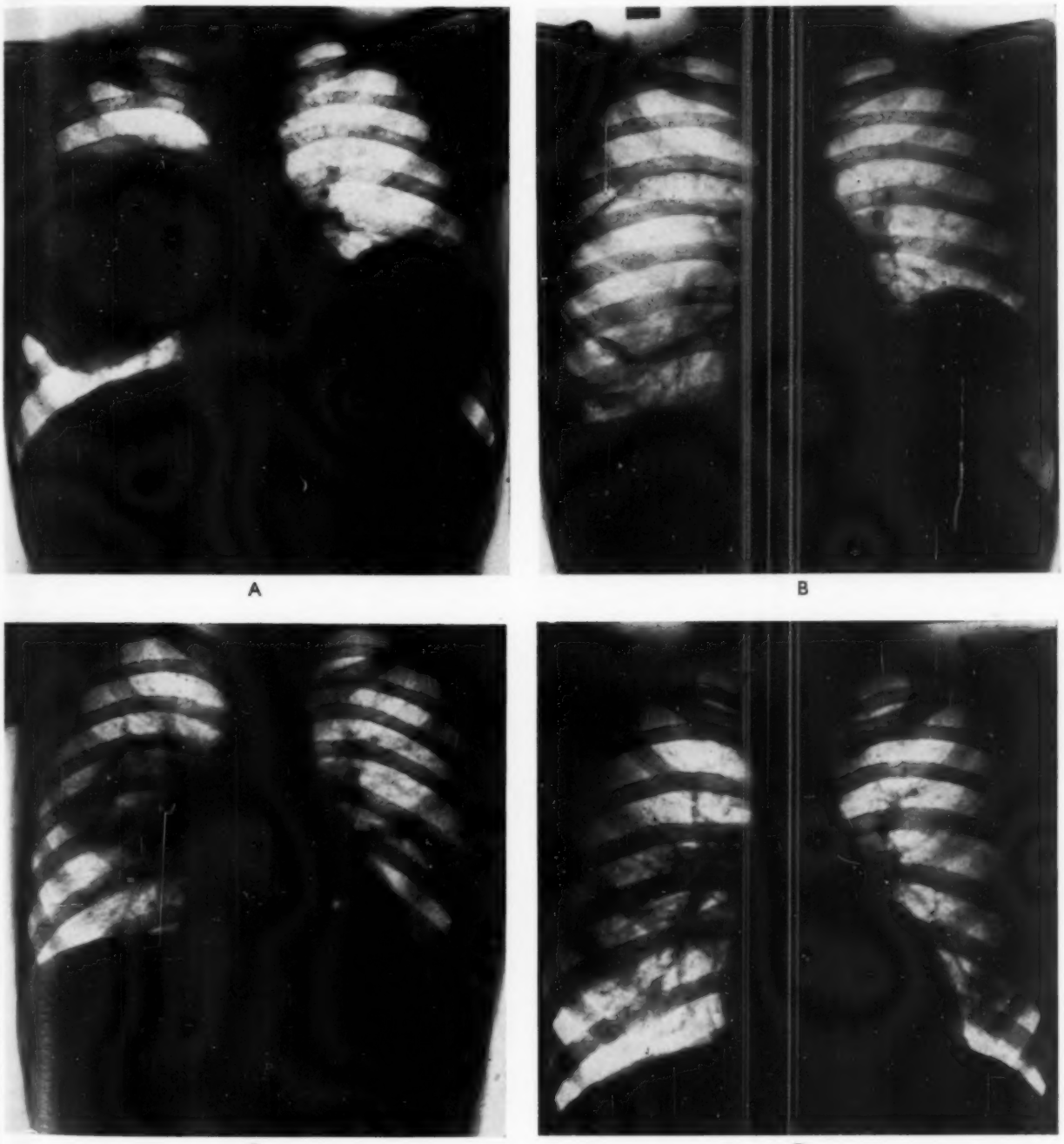


FIG. 295.—These four films show the fate of the 'empty sac' in a child, aged 6, who suffered from bilateral simple hydatid disease. A, The first film shows the bilateral cysts. B, The hydatid in the right lung has been removed and the 'empty sac' remains. No attempt was made at operation to obliterate the space after taking the parasite out. C, The hydatid on the left has now been removed, six months after the first operation. The original sac is still present on the right and the new one is also shown on the left. D, This film was taken 18 months after the first operation. Both sacs have now disappeared. The child was clinically normal.

left. (*Fig.* 295.) Subsequently both sides were shown to be normal by bronchography. In this respect the empty sac resembles the large bubble shadows which are characteristic of healing staphylococcal pneumonia, or the ' ring ' shadows of tuberculosis. As long as the sac is seen on radiographs to be empty it can be assumed that it is sterile ; it probably remains

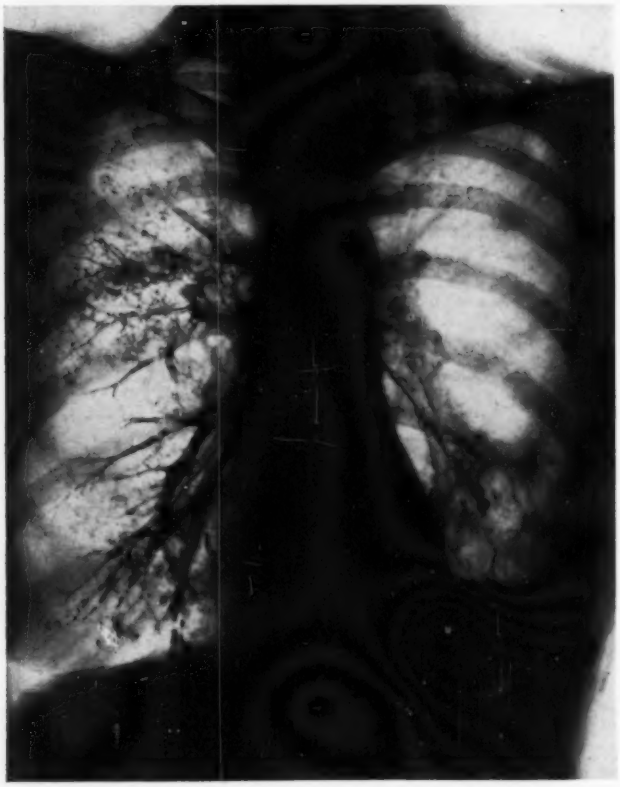


FIG. 296.—This bronchogram shows lipiodol in a persistent empty sac at the left base. The cavity outlined is the original cavity in which the parasite grew, and, being completely peripheral and dependent, it has not obliterated spontaneously. It was not causing symptoms and was thus not treated surgically. It may cause trouble in the future.

sterile because the cavity is not primarily bronchial in origin but is a lateral diverticulum in the interstitial tissue of the lung. The diverticulum connects with the bronchial tree by a slit-like valvular opening and hence it is not likely to become infected or to fill with mucus.

In recent years surgeons, following up their cases of hydatid disease after removal of the parasites, have made bronchograms and produced evidence that in a significant number ' bronchiectasis ' can be demonstrated, even when it has not been expected. (*Fig.* 296.) This finding has led some to advocate lobectomy as the treatment of choice for pulmonary hydatids. We now believe the facts to be as follows. In most cases the residual space obliterates spontaneously and quickly, and causes no further trouble ; this happens almost irrespective of how it has been treated at operation. In a minority a tension cyst, which ultimately becomes lined by epithelium, forms and this may give rise to the signs or symptoms of an ordinary lung cyst ; on the other hand it may persist in a quiescent state. This lesion is a cyst and it should not be called ' bronchiectasis '. But true bronchiectasis can occur, and is sometimes present at the time the parasite is enucleated. It is due to bronchial obstruction by the hydatid. These cases are rare ; but if bronchiectasis is present at the time the parasite is first treated one can make out a case for lobectomy or pneumonectomy to avoid trouble later on. (*Fig.* 297.)

In the present series residual cavities have been demonstrated on several occasions but have not occasioned the patients any trouble. Nevertheless we feel that efficient treatment demands more than simply taking the hydatid cyst out of the lung and to deal effectively with the ' empty sac ', one of two procedures may be used.

If the original cyst was small, the bronchial fistulæ should be closed individually, and the ' sac ' obliterated by a series of purse-string sutures inserted *seriatim* from the bottom to the surface of the space. If, on the other hand, the original cyst was large and has been enucleated successfully, then it is wise to excise the adventitia and to close the main segmental bronchus.

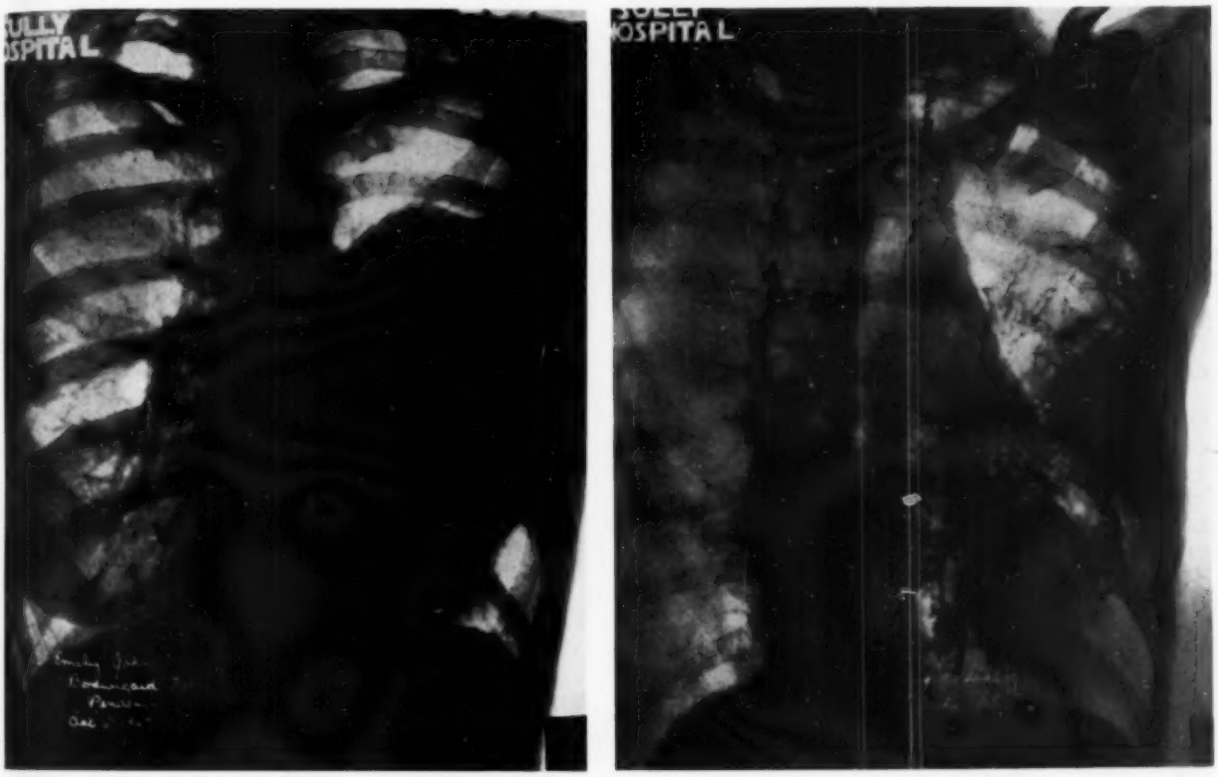
Concerning the Actual Methods of treating Simple Hydatid Cysts used in this Series.—The treatment of simple hydatid cysts in the lung has varied in this series according to increasing familiarity with the details of the pathology of the lesions and the progress of thoracic surgery in general. Moreover, some of these patients were operated upon on the assumption that they were suffering from some condition other than hydatid disease, and the correct diagnosis was sometimes not made until after the abnormal parts had been removed. Thus all the operations done were not necessarily the best suited to the individual case.

Most of the various techniques advocated by others have been tried, but we now aim at *removing simple hydatid cysts from the lung intact*. This operation we have practised successfully on 17 occasions and in 4 others we have tried to remove the cyst in this way but have ruptured it in the process. At these attempts the possibility of the cyst bursting has always been in mind and the wound has been packed off and suction has been available so that the patients were not worse off than if they had been treated by other ways. The results in all these patients were excellent, and in none were there any serious post-operative complications.

Eight simple cysts were treated by *lobectomy* and one by *pneumonectomy*. These operations were either done because the pre-operative diagnosis was in doubt or because the hydatid was so large that at time of operation it was considered likely to have destroyed the lobe in which it lay. This reasoning was probably wrong and, in future, most cases of this type will be treated by removing the parasite intact. All but one made a good recovery and there were no difficult or serious post-operative complications. One patient who had a pneumonectomy done (C.P.T.), on the assumption that the lesion was a carcinoma, died as a direct result of operation.

On twenty-seven occasions simple hydatid cysts were treated by deliberate *thoracotomy*. The presence or absence of pleural symphysis was ignored ; the cysts were taken out of the lung after aspiration ; the empty sacs were obliterated as well as possible, and

the pleural cavities were drained. These cases were not free of complications. One became infected and had an empyema drained, one developed a severe tension pneumothorax and in one the pleural cavity filled with blood and a decortication was necessary to avoid a 'frozen chest'. All the 27 patients appeared to do well ultimately but 4 developed daughter cysts later.



A B

FIG. 297.—A, Radiograph of a woman's chest. It shows a large hydatid cyst in the left lung. The patient had the signs and symptoms of bronchiectasis and so a bronchogram was made. B, The bronchogram shows that the hydatid has ruptured in the interim and that the patient has gross bronchiectasis; the treatment was pneumonectomy and after the operation the cyst was found to be infected. A good clinical result has been achieved.

In 13 cases of simple hydatid cyst a *two-stage operation* with marsupialization of the lung, or if existing pleural adhesions were present, a one-stage drainage, was done. Two of these patients developed daughter cysts and in 3 convalescence was complicated by an empyema. In one of the latter the empyema became chronic and the patient died on the operating table when an attempt was made to obliterate the cavity later.

There has been divergence of opinion as to whether, in man, it is harmful to contaminate a serous membrane or an operation incision with liquid from a hydatid cyst. The cases in this series have proved that although the immediate results appeared to be satisfactory in this group there were 6 patients out of 40 who developed daughter cysts or secondary echinococcosis. This incidence is lower than we had anticipated but high enough to be a sound argument against spilling hydatid fluid. Moreover, there is another reason to avoid bringing hydatid fluid into direct contact with the host. Only one of the patients in this series developed serious post-operative symptoms due to anaphylaxis. The question may be relevant not only to the operation in hand, but, if the patient harbours other parasites, to any operation for hydatid disease which may be required later on; and in this connexion it is generally believed nowadays that a general anæsthetic offers doubtful protection against anaphylaxis. Thus, if the wound has been contaminated and if a second operation is necessary to remove a second cyst it is wise to desensitize the patient by injecting increasing doses of hydatid fluid.

The Treatment of Infected and Ruptured Lung Cysts.—In some cases the correct diagnosis will not have been made before operation and the patient will be considered to be suffering from chronic pulmonary suppuration, tuberculosis, actinomycosis, or carcinoma. If a definite history is not available to suggest hydatid disease, the diagnosis is only likely to be suspected in the British Isles if fragments of laminated membrane have been expectorated or if they have been seen at bronchoscopy. This is fortunately not vital to the patient for the presence of a chronic lung abscess demands active surgical treatment.

The problem is to cure a patient suffering from a special type of chronic lung abscess in which there is a persistent sequestrum. That sequestrum consists of the disintegrating remnants of the laminated membrane of the parasite; or, possibly, of a number of daughter cysts living meagrely in a collection of bloody pus and debris.

The modern conception of the treatment of chronic lung abscess is lobectomy. This treatment is effective and cures the patient of an ailment which, if left undisturbed, progresses to diffuse pulmonary suppuration. It is based upon the pathological and histological examinations which have been made upon the tissues removed at operation, and these have shown that an established, chronic, lung abscess totally replaces the normal lung tissue, in which it lies, by scar, inflammation, and suppuration. A lung abscess due to hydatid disease behaves somewhat differently in that the spread of the inflammatory process, far and wide beyond the adventitia, only takes place slowly and in many of these cases a chronic hydatid abscess can exist for a long time without destroying everything around it. On the other hand, the end-result of the process is the same and some of the long-standing hydatid abscesses in this series had not only destroyed the lobe which confined them, but the tissues were firmly adherent by scar and inflammation to everything in the neighbourhood. In fact removal of a hydatid abscess can be a most difficult operation.

The practical point about these observations is that whereas lobectomy will be the method of election in the management of many cases, a few can still be cured by incision and drainage of the abscess, with removal of the necrotic contents. The patients who are most likely to respond to drainage are young people whose radiographs and physical signs suggest that the process is still localized to one part of the lung; and those whose general condition is good and who have little or no expectoration. The final decision as to the method of choice—as between lobectomy and drainage—can only be taken in most instances at operation; but wherever there is doubt as to the wisdom of drainage lobectomy should be done. The operation may be very difficult indeed but the results are final and excellent.

The treatment of daughter cysts has been more difficult than that of simple hydatid disease, and the reason has been that the laminated membrane in these cases does not seem to be strong, and it is very difficult to get the parasites out intact. Thus in most of these patients a lobectomy has been preferred to enucleation. The complex situations which may arise are illustrated in the following case.

A young man was admitted to St. Thomas's Hospital and the changes which had occurred in his lung were as follows. A radiograph, taken 3 years previously, showed a solitary and simple hydatid cyst in the right lower lobe. This ruptured spontaneously and within a year a number of shadows began to appear in the right lung. It was obvious that the original cyst had re-established itself and since he had the clinical signs of a lung abscess it was probable that infection had occurred. At thoracotomy four different groups of cyst were found in the right lung. The original hydatid had grown again, it was uninfected and there were two daughter cysts. At the back of the lower lobe was a simple cyst the size of a tangerine. In the upper lobe there was a simple cyst the size of a pea and in the middle lobe there was a hydatid abscess, full of pus and containing impacted membranes. This patient was treated by removing all the parasites, and closing the abscess, having aspirated the sequestra of laminated membrane and leaving antibiotics in the space. It was not possible to do these various manipulations without bursting two of the hydatids.

This case raises the possibility of bronchial dissemination after rupture. It seems difficult to explain the presence of such scattered cysts on any other basis.

The Methods used to deal with the Infected Cysts in the Present Series.—In 12 cases the cysts were treated by opening the abscess, and removing the sequestra. In this group there were dense pleural adhesions whenever the abscess was situated near to the parietal pleura; but when it lay at some distance from the surface of the lung the pleural cavity was either free or there were limiting interlobar adhesions. Two different techniques were used to meet these circumstances. When drainage had been decided upon and adequate localizing adhesions were found the abscess was drained in the usual way; but in some instances there were no pleural adhesions and then the abscess was opened 'across the pleural cavity', the debris was removed, antibiotic powders were left in situ and the cavity in the lung was sewn up—the reasoning being that it was possible that the lung cavity might obliterate forthwith without suppuration or empyema, and that if these complications did develop they could be dealt with in due season. This technique worked without a hitch in the 3 instances in which it was used. There were no deaths in this series; 12 infected cysts were successfully treated by lobectomy and 3 by pneumonectomy.

FOLLOW UP

The 71 patients reported in this paper have been treated between the years 1938 and 1952, and as far as we have been able to ascertain only 7 are unsatisfactory at the present time.

1. A man, aged 40, who was judged to be suffering from carcinoma of the bronchus underwent pneumonectomy and died soon afterwards. The lesion was a hydatid abscess situated in the upper lobe, and the dissection of the matted tissues was so difficult that hæmorrhage and shock occurred. This is the only operative death in the series.

2. A girl, aged 11, had a large, simple cyst removed by thoracotomy and aspiration from the right lung. The operation was done in August, 1940. Her convalescence was uneventful and she remained well until 1946 when she developed a 'tumour' in the right iliac fossa. This proved to be a hydatid projecting from the under-surface of the liver. She underwent laparotomy. In 1947 multiple hydatids were again removed from the liver and the pelvis; in 1949 two more cysts were taken from the liver and in May, 1951, she was found to have 2 cysts in the pelvis. She is alive to-day.

This patient was not known to harbour abdominal parasites whilst she was in Sully Hospital and there has been no evidence of recurrence in the thorax.

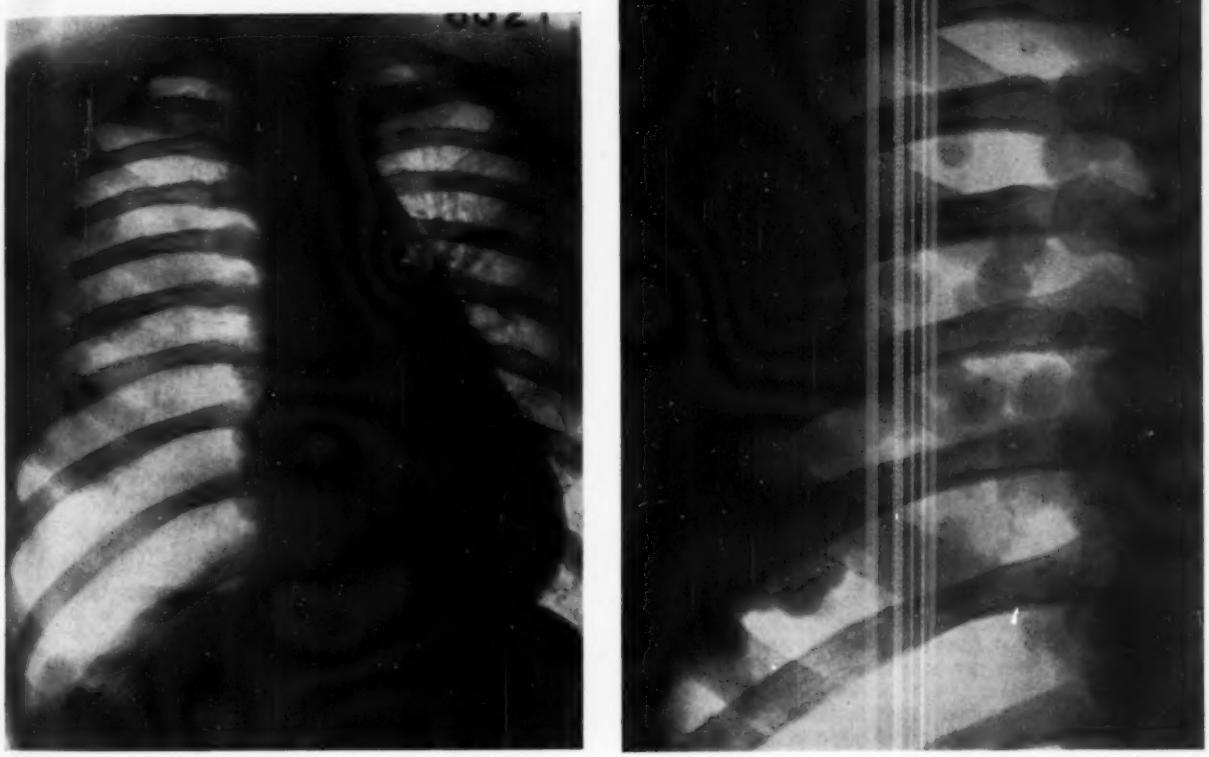
3. A woman, aged 50, came up complaining of empyema. The empyema was due to a hydatid cyst which had herniated out of the lung, ruptured, and produced multiple secondary cysts in the pleural cavity. The empyema was drained and out of the drainage-tube came large numbers of small, live hydatid cysts and tubercle bacilli. The empyema did not resolve and the patient died of tuberculosis two years later.

4. An Italian prisoner-of-war developed large numbers of daughter cysts in the pleural cavity after a pulmonary hydatid had herniated out of the lung and burst in the pleura. (*Fig.* 298.) This patient was repatriated without

treatment being attempted ; but faced with a comparable problem to-day one would probably treat the condition by pleuropneumonectomy. With care it should be possible to get such a lung out and to keep the pleura as an intact bag containing the daughter cysts.

5. A woman who developed an empyema after treatment died as a result of an attempt to cure the empyema surgically.

6. One patient was first seen just before death. There were two cysts in one lung and one in the other. One



A B

FIG. 298.—A, Radiograph of the chest showing the state of the patient on admission. B, A magnified photograph of some of the pleural cysts. An Italian soldier, aged 23, was admitted to hospital, January, 1943, having 6 months before become ill with 'right basal pneumonia and pleurisy'. A radiograph at this time was stated to show a right-sided resolving empyema. Five months prior to admission suddenly developed a right tension pneumothorax, which persisted in spite of numerous withdrawals of air. Small quantities of clear sterile fluid were also aspirated. On admission the patient was dyspnœic, with signs of right pneumothorax with gross mediastinal displacement to the left. Thoracoscopy revealed numerous spherical translucent cysts lying scattered about on the visceral and parietal pleuræ, and on the diaphragmatic surface was a large collapsed laminated membrane, apparently the parent cysts which had ruptured into the pleura liberating daughter cysts. (Subsequent history of case not known.)

was simple, one had ruptured, and one had ruptured and was infected. The patient died before any treatment could be instituted. (*Fig.* 299.)

7. A man, aged 21, had a lobectomy successfully performed in 1942 for a large hydatid in his right lower lobe. On sectioning the lobe it was found to contain scattered tubercles which were active. He made an uninterrupted convalescence and remained well for 9 years, during which time he has continued to work on a farm and has since developed a hydatid cyst in his liver, which has been successfully removed.

Apart from the cases described above the remainder have been cured of their hydatid disease. But this statement would convey a wrong impression if it suggested that almost any surgical treatment is safe and adequate.

One of the most striking features has been the serious post-operative complications which occurred as a direct result of imperfections of technique at the time of operation. In some of the early cases, when it was customary to rely on, or to create, pleural adhesions, to hold the lung to the chest wall after the parasite had been removed, acute tension pneumothorax and empyema occurred. These complications were corrected and overcome ; but they added to the hazards and prolonged the discomforts of convalescence. They could kill patients ; indeed one of the patients described above died as a result of an attempt to obliterate a chronic empyema surgically. Nevertheless recent advances in thoracic surgery have made the management of pulmonary hydatid disease almost safe, and most authorities claim that the operative mortality should be less than 1 per cent.

The fact that daughter cysts or secondary hydatid disease ultimately occur in at least 10 per cent of cases in which the cyst fluid has been spilt at operation, is a strong point in favour of doing an operation which avoids this possibility. The experience afforded

by the present series of cases is that this cannot be achieved with certainty by aspirating the cyst before 'removing the membranes intact' — because the membranes are not then intact. Hence we advocate enucleation of the parasite, without aspiration, according to the techniques already described. Using this technique, there have been no serious post-operative difficulties and no cases in which recurrence has taken place. But one of the unexpected results of this investigation has been to show that even if the lung cavity, the pleura, and the chest wall are contaminated by rupturing the cyst, recurrence of the disease will not always occur on this account. The actual risk has been to 6 patients out of 40.

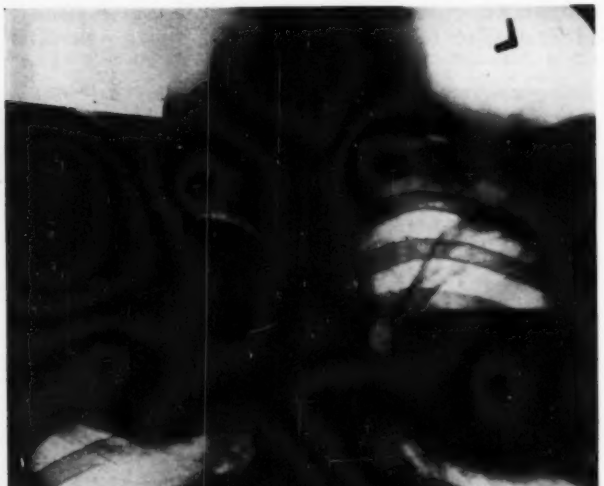


FIG. 299.—Radiograph of a patient who died, untreated, from hydatid disease. One cyst, in the right upper lobe, shows a perivesicular pneumocyst: that in the right lower lobe is a simple hydatid cyst and the one in the left upper lobe is a hydatid abscess.

Lobectomy and pneumonectomy have produced cures in so far as the hydatid disease has been concerned.

Anaphylactic reactions may have occurred in this series of patients, but it has never been easy to differentiate between signs and symptoms due to this cause and those due to other reasons. There have been occasions when a patient has suffered from shock, tachycardia, dyspnœa, etc., which were not well explained by the physical findings; but who is to say whether these were manifestations of anaphylaxis or not?

The Casoni reaction has generally proved to be reliable both in simple and complicated cysts; but the difficulties associated with getting fluid which is certainly active, and of interpreting the test, are such that it does not occupy the principal place in diagnosis. Moreover, there has been a small group of patients, seen during the same period, in which a positive Casoni was obtained but the patients were not suffering from hydatid disease. The commonest cause of false positives has been in patients suffering from malignant disease and especially in those who have multiple secondary deposits. The Casoni reaction does not change quickly or certainly from positive to negative after removal of the parasite, and it may still be positive five years or more in a patient in whom there is no evidence of persisting infestation. Thus the reaction cannot be used either as a guide to cure of any specific lesion, or to indicate the presence of other hidden cysts.

Acknowledgment.—The authors are indebted to Sir Clement Price Thomas not only for permission to scrutinize his cases, but for his constant help in all things.

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ACTINOMYCOSIS OF THE TESTIS

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ACTINOMYCOSIS rarely affects the genito-urinary system and then as a rule it involves only one organ.

Nevertheless, for many years this condition has been recognized in the kidney. Edgar Garceau (1909), in his book entitled *Tumours of the Kidney*, discusses the findings in 128 autopsies on cases of actinomycosis; in 11 (8·6 per cent) one kidney contained metastatic abscesses, in a further 11 cases the kidney was involved by continuity with adjacent organs, and in only 2 was the disease apparently primary in the kidney. He mentions no case in which the testicle was attacked.

Reviewing actinomycosis in the United States of America, Sanford and Voelker (1925) report only 8 instances of genito-urinary involvement out of a total of 670 cases investigated; in only one case was the testis affected. In this review the precise location of the lesion is often not stated, so that the relative incidence of genito-urinary actinomycosis is uncertain.

Examples of renal infection are recorded from time to time and emphasis is laid on the difficulty in diagnosis. Instances have also appeared of involvement of the ureter and bladder (secondary to

an adjacent lesion), and of the scrotum and penis (by invasion from outside), but these must be very rare.

Only 4 examples of actinomycosis of the testis have been discovered; in none of them was the diagnosis made before operation. Acute and chronic inflammatory disorders of the testis are common, so it is, perhaps, surprising that the organ is not more often attacked.

The four cases reported in the literature are considered and a fifth, diagnosed before operation, is presented.

CASE REPORTS

Previous Examples of the Disease.—

Case 1.—This was reported at a medical meeting in Sussex by Dr. A. Eustace in 1899. A man of 35, 'a gentleman of independent means', visited his doctor with a swelling in the left cord which he had noticed for three months. "Examination showed (1) a hard cylindrical swelling, painless and not tender, on the left cord, about the size of a hazel nut, and situated about one inch from the external abdominal ring; (2) a small hydrocele of the cord; (3) general enlargement of the testicle but chiefly of the globus minor; no enlarged glands; rectal examination and base of bladder normal." A diagnosis of a chronic inflammatory process "probably tuberculous" was made and operative treatment advised.

Mr. Victor Horsley of London saw the patient in consultation five days later and supported the practitioner in his diagnosis and treatment. An orchidectomy was carried out, the cord being divided half an inch above the swelling; the wound healed by first intention. Potassium iodide gr. 3 t.i.d. was prescribed for three months.

The operation specimen was sent to Mr. Horsley, who examined it microscopically. The section "exhibited the typical appearances of *Actinomycosis bovis* and the result of its presence in the tissues". The speaker said that this was probably the first recorded case of actinomycosis of the testicle. The report was made 16 weeks after the operation, when the patient was very well and had increased 1 st. in weight.

Case 2.—In the survey of actinomycosis in the United States of America in 1925 (q.v.) the following information is given about a case involving the testicle. A man of 47, a minister, suffered from an inflammatory condition of one testicle thought to be due to tuberculosis. The symptoms were of ten months' duration. After orchidectomy the condition was found to be due to actinomycosis. No other clinical details are available.

Case 3.—This was discovered incidentally (Schneider, 1945). A man of 80, a farmer, was admitted to hospital with a urinary retention of four days' duration, a catheter having been passed every 8 hours to relieve him. He was treated with an indwelling urethral catheter for a further three days; a suprapubic cystostomy was then performed. Twelve days after the onset of urinary retention he developed a right epididymitis with a fever of 106° F. The author then states "this was treated in the usual manner, and subsided rapidly".

About 3½ weeks after his admission a bilateral vasligation and transurethral resection of the prostate were done. Adenocarcinoma of the prostate was diagnosed. Four weeks later a left subcapsular orchidoplasty and a right orchidectomy were performed. The clinical improvement was rapid and the patient was discharged home fit with a healed suprapubic wound and normal micturition. He was alive and well 3½ months later.

Section of the right testis showed "nodular areas of pale grey and yellow tissue occupying the peripheral half of the testicle. Nodules are confluent, but measure individually 2 mm. in diameter". A diagnosis of actinomycosis was made on microscopical examination of the testis; the epididymis was normal.

Case 4.—Here also the discovery of the disease was incidental (Baker and Ragins, 1946). A man of 77, suffering from carcinoma of the prostate with secondaries in the lumbar spine, was treated by endoscopic resection and 9½ g. of tissue were removed. Thirteen days later "an acute bilateral epididymitis developed which subsided rather rapidly". After six weeks a bilateral orchidectomy was performed. Actinomycotic abscesses were found in the right testis, the cut surface of which was "brownish grey and studded by numerous abscesses which were filled with a greenish grey purulent material emitting a musty odour". The epididymis was normal to naked-eye inspection. Microscopically, actinomycotic granules were found embedded in many of the abscesses; the epididymis was not inflamed.

The patient made an uninterrupted post-operative recovery apart from a transient purulent discharge from the wound on the right side of the scrotum; no fungi could be identified in the discharge.

A further resection of prostatic tissue was performed six months after the first, but the patient did not survive this by more than a few weeks.

A Current Case Record.—

Case 5.—

HISTORY AND CLINICAL FINDINGS.—A man, 52 years of age, a tailor's cutter, presented as an out-patient in April, 1947, saying that he had noticed a swelling of the left testicle during the preceding six or eight weeks. It was slightly painful and was increasing very slowly in size. He had suffered no injury, had no urinary symptoms, and in all respects he considered himself in good health.

His past medical history was uneventful except for prolonged intermittent dyspepsia. A perforated duodenal ulcer had been repaired in 1940.

Examination revealed a swelling the size of a duck's egg in the left side of the scrotum. It was uniformly hard except for a small area at the upper pole, which was fluctuant. There was no tenderness on palpation and the normal testicular sensation was present. The epididymis could not be defined as a distinct structure and the cord was thicker than normal. The scrotal skin was adherent anteriorly, but it was not inflamed. There was no inguinal hernia, and no enlargement of either abdominal or inguinal lymph-glands could be detected. Rectal examination revealed a prostate of size average for his age; the vesicles were not palpable.

A possible diagnosis of testicular neoplasm was made and further investigations were carried out. A chest radiograph revealed normal lungs. His W. R. was negative.

COURSE OF THE DISEASE.—During the next week the fluctuant area increased in size and on May 6, fluid was aspirated so that the bacterial content could be examined. The few cubic centimetres thus withdrawn showed small white granules which consisted of finely beaded filaments with fine branching typical of actinomycosis. Aerobic and anaerobic cultures maintained for seven days remained sterile.

On the following day a profuse discharge of thin, brown, offensive fluid was taking place from an anterior sinus. The skin was now reddened and swollen, but the testicular swelling remained the same size. He was admitted to hospital forthwith, rested in bed with a scrotal support, and given large doses of penicillin parenterally. After five days there was no change in the condition of the swelling. His general well-being remained good and he had no rise of temperature.

OPERATIVE TREATMENT.—On May 19, twelve days after his admission to hospital, there being no material

change in the size of the testicle or amount of discharge, an orchidectomy was performed by Mr. H. G. Hanley. The cord was removed up to the internal ring and a substantial area of scrotal skin around the sinus. The wound was closed around a drain, which remained in position 48 hours. Healing was slow and when he left hospital on June 2 there was still a discharge of thin seropus. Within a further two to three weeks healing was complete.

PATHOLOGICAL REPORT.—" The specimen consists of the left testis with adherent envelopes and an area of dark-brown scrotal skin surrounding a sinus, the whole

FIG. 300.—Actinomycosis of the testis. The appearance of the organ on cross-section, showing the honeycomb appearance of the disease and an anterior fistula.

constituting a firm indurated mass. The cut surface shows the outline of the testis involved and almost surrounded by confluent fibrocaseous areas of a yellow and white colour. The general appearance suggests tuberculosis. (*Fig.* 300.)

" Histological examination reveals a fibrino-purulent inflammation in which scattered colonies of actinomyces are readily identified (*Fig.* 301).

" No areas suggestive of tuberculosis have been found in any of the sections."

SUBSEQUENT HISTORY.—There has been no further evidence of actinomycosis in the patient. In November, 1949, a partial gastrectomy was performed for duodenal ulceration by Mr. Marnham, at St. George's Hospital. Mr. Marnham states that, as far as can be told, there was no evidence then of hepatic or intra-abdominal actinomycosis.

In September, 1950, he presented at hospital with an influenzal pneumonia ; opportunity was taken of carrying out a full urological examination and no sign of disease was detected. The orchidectomy scar was sound. His good health continues at the time of writing.

DISCUSSION

Aetiology.—Actinomycosis in man is caused by an anaerobic organism, *Actinomyces bovis*, which is commonly present in the oral cavity where it remains latent. Entry into the tissues is achieved only after the integrity of an epithelial surface is impaired by disease, accident, or surgical operation (Cope, 1947). A local reaction then occurs and is usually subacute or chronic in nature.

Small abscesses form, surrounded by a hard fibrous tissue. Extension takes place commonly by direct continuity, but from time to time cases occur in which the organism gains access to the blood-stream and a metastatic focus occurs in a situation remote from the primary. If the primary focus is small and becomes quiescent or heals, it may be that the first manifestation of the disease is at the site of the metastasis. In such a case this lesion bears the name ' primary '.

Source of Infection.—When an actinomycotic lesion occurs in the kidney in the absence of any evidence of the disease, blood-borne infection from a small quiescent or healed focus elsewhere, possibly the lungs, is presumed. In actinomycosis of the testis some similar explanation may be given for the source of the disease. But other suggestions can be entertained in considering the five cases recorded above.

In *Case* 4 the writers state that the infection may have been blood-borne and have arisen in carious teeth. Nevertheless it is interesting to note the onset of an epididymitis six weeks before orchidectomy and to speculate on the likelihood of the organism being implanted in the posterior urethra either by the passing of an instrument from below or by the urine coming from above. The same observations apply in *Case* 3, where a bilateral

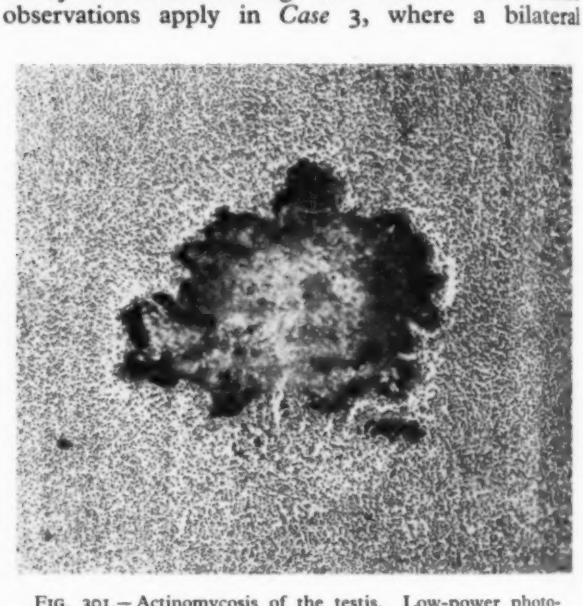

FIG. 301.—Actinomycosis of the testis. Low-power photomicrograph, showing the typical ray fungus. (× 60.)

epididymitis followed the passage of an instrument and actinomycotic abscesses were found in the testis six weeks later.

Since actinomycosis spreads most easily by direct invasion, and is particularly prone to do so within the bounds of the abdomen, extension of the disease down the cord to reach the testicle is a not unlikely supposition. In the case recorded by Dr. Eustace (*Case* 1) a nodule was present in the cord above the diseased testicle. This may be evidence of the descent of disease from above. That the man was alive and well twelve weeks after the operation does not disprove the possibility of a primary intra-abdominal focus of the disease, but against it is the fact that actinomycosis is much more common on the right side of the abdomen and the patient's left testicle was affected. It may equally be argued that the disease spread from the testis up the cord, or that the nodule in the cord was caused by some agent other than the streptothrix.

Although renal actinomycosis in some respects resembles renal tuberculosis, no case of associated

epididymitis or orchitis has been recorded in the former condition, but when it is considered that actinomycosis rarely spreads in the lymphatics (Cope, 1947) and tuberculosis commonly does, the fact is not surprising. Involvement of the testis by way of the perivasal lymphatics is therefore not to be expected in actinomycosis.

On the whole, spread of the disease in the blood-stream to reach the testicle is most likely. It is known that actinomycosis can enter and pass through the body, leaving behind no trace of its passage; a small primary focus occurring in the lung or bowel heals completely. In the stage when the primary infection is active metastasis takes place, but it is only the secondary lesion that becomes clinically evident. It is presumed that this is so in *Case* 5 and the lapse of five years without demonstration of disease elsewhere in the body suggests that it actually took place. *Case* 1 may also have been one in which the infection was blood borne. In *Case* 2 there are insufficient details to enable any opinion to be given.

Several examples of primary actinomycosis of the scrotum have been recorded, but, as far as is known, in none has the underlying testicle been affected.

Symptoms.—The earliest clinical description of the disease applies with astonishing accuracy to the case which prompted this study (*Case* 5), although it was written of the disease as it affects the face and neck. J. Israel (1886), in his excellent account, says, "Wherever the point of origin may be, actinomycotic tumours at first feel hard, and do not affect the skin. Their course in strictly typical cases is never an acute inflammatory one, but slow and torpid, extending usually over several months, and scarcely exciting any attention by reason of painfulness. Pain is usually not felt until after some time, and after changes have taken place in the tumour. It is only quite exceptionally that any general symptoms are present in the class of cases under consideration. Secondary changes may occur, consisting at first of false or true fluctuation in the tumour, according to the amount of fluid it contains. The fluid is in most cases thin, seropurulent, and odourless, and contains the characteristic granules. If the process goes on undisturbed discharge will take place through the skin; and though the openings may in rare cases close, they most frequently continue as fistulæ with a slight discharge."

Clinically, therefore, the gradual onset, the hard painless tumour, its softening and fluctuation in one place, with the subsequent formation of an unhealing fistula, are all features to be expected, as well as the lack of systemic disturbance. The superficial position of the testis, of course, allows for the early formation of a fistula.

Diagnosis.—In the diagnosis of actinomycosis Cope (1949) gives three reasons for the disease being overlooked: there is no reliable reaction or blood test for the disease; it may be a long time before an abscess forms from which organisms can be obtained; and the diagnosis may not be considered. In a case of subacute or chronic inflammatory orchitis of obscure origin the possibility of actinomycosis should not be overlooked. Certainty can only be achieved by finding actinomyces in the pus or the tissues of the affected area; bacteriological examination of any discharging fluid should therefore be made and histological examination of accessible tissue not be neglected if bacteriological findings are inconclusive. Also, if in all cases in which the testis is removed for chronic inflammatory disease a similar careful examination is made, other cases of actinomycosis may come to light.

Treatment.—The treatment of the disease consists in simple orchidectomy. To quote Israel again: "The danger arising from actinomycosis depends chiefly on the locality where the disease is situated, the importance of the organs affected, and the greater or less facility for getting at it for operation". The disease as it affects the testicle is readily accessible, and can be removed without danger to health assuming the opposite testicle to be normal. If the disease is in fact 'primary' the prognosis is excellent.

The case recorded above did not respond to the use of penicillin; a course of five days was given, with a dosage of 50,000 units 3-hourly. A more prolonged and more intensive course might have been effective, as it has been in examples of the disease occurring elsewhere. The newer antibiotics may in time prove their worth.

SUMMARY

A brief survey is made of actinomycosis of the testis. Four cases previously reported in the literature are reviewed and a fifth is added.

I am indebted to Mr. H. G. Hanley for permission to publish this case; to Dr. H. Rogers for his pathological report and his advice; and to Miss F. Wadsworth for her drawing.

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ILEOCÆCAL ARGENTAFFIN CARCINOMA WITH DISTANT METASTASES

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ARGENTAFFIN carcinoma of the alimentary tract is not uncommon, occurring especially in the appendix. In the ileum it is found about half as often as in the latter, and more rarely in the rectum, stomach, duodenum, jejunum, colon, and cæcum. It has also been described in the gall-bladder, intestinal mucosa in ovarian teratomata, pancreas, and in the bronchus, though in the last two sites there is some doubt as to the true identity of the tumour. From time to time cases with metastases occur, but unless all such cases are reported, however shortly, the true incidence of these will never be known. Up to the beginning of 1951, just over 100 metastasizing argentaffin carcinomas were described in the literature, mostly occurring in the ileum.

At this hospital, during the five years from 1946 to 1950, 980 appendices have been examined histologically and, in these, 4 cases of argentaffin carcinoma were discovered (0·41 per cent). Figures ranging from 0·2 to 0·5 per cent are quoted in the literature. A figure of 0·28 per cent was given by Porter and Whelan (1939) for a series of 26,384 appendices examined from 1910 to 1937. In this same period they found 11 cases in other parts of the alimentary tract, 6 of which were in the ileum. Of the latter, 3 were with metastases, one being removed surgically with no recurrence ten years later and one being found at operation for obstruction, with death of the patient some days later. The third example was found incidentally at autopsy. In the first and last cases spread was limited to the regional lymph-nodes, but in the other case there was widespread dissemination to the liver, pancreas, adrenals, and vertebræ in addition. Willis (1948) refers to 8 cases of small-bowel argentaffin carcinoma, 4 of which had metastases to distant organs. Details of 6 of the cases were reported in 1940 (Willis, 1940), with a discussion on malignancy and metastasis in this tumour. Dockerty and Ashburn (1943) recorded 13 further examples.

In the author's department argentaffin carcinomas have been found 3 times in 1590 autopsies during the past 5 years (0·18 per cent), compared with a figure of 0·34 per cent for 2922 autopsies by Porter and Whelan (1939) and from 0·25 to 1·8 per cent for other authors. Obviously the more assiduous the search the greater would be the frequency. In the surgically removed appendices at Ashford Hospital no evidence of metastasis has been found, and this was also true of Porter and Whelan's cases. They suggested that argentaffin carcinomas arising in fibrosed appendices may be proliferations due to chronic inflammation and not true neoplasms, but another possibility for the rarity of metastases is that in the appendix the condition usually gives rise to early symptoms and there is often not time for metastasis to develop. The age incidence of the appendiceal tumours is greatest in the third decade, but in the fifth decade or later for those in the small bowel. However, argentaffinomas of the ileum sometimes give rise to obstruction quite early, yet metastases are frequent. In a series of 152 'carcinoids' of the small intestine in the literature, 37 had metastasized according to Humphreys (1934). It appears, therefore, that small-bowel argentaffinomas have a greater tendency to metastasize than those elsewhere. A case has been seen here in which acute ileo-ileal intussusception was caused by an argentaffin carcinoma half the size of a pea. This was removed and the patient is well two years afterwards. If the primary tumour is removed together with any removable metastases which may be present, the prognosis is not as bad as that in adenocarcinoma (e.g., the cases of Stewart and Taylor, 1926; Cope and Newcomb, 1930; Porter and Whelan, 1939). In the ileum these tumours tend to occur at the lower end; they are often multiple, and this is a point of some importance in assessing prognosis.

CASE REPORT

Mrs. E. G. S., aged 85 years, was admitted to Ashford Hospital with multiple bruises after a fall. The following day she complained of pain in the right iliac fossa and diarrhœa. On questioning, it appeared that the latter had been present for about six weeks, without blood or mucus, and that there had been a rather rapid loss of weight, but a good history was difficult to obtain.

FIG. 302.—Photograph of specimen. (× ⅔.)

ON EXAMINATION.—The patient was thin, dehydrated, and anæmic. A tumour about an inch in diameter was felt in the right iliac fossa, which was tender and less mobile up and down than laterally. The liver was slightly enlarged, with a hard irregular edge. *Per rectum* a hard irregular fixed mass was felt 5 in. from the anus. A diagnosis of carcinoma of the cæcum and diverticulitis of the pelvic colon was made. A barium meal showed diverticulosis of the colon but revealed no neoplasm in the cæcum. Tests for occult blood were positive in the second of three consecutive daily stools. Blood examination

revealed an iron-deficiency anæmia, with a hæmoglobin of 9·9 g./100 ml. (67 per cent). There were no symptoms of obstruction and operation was not thought advisable. The patient's condition gradually deteriorated and death occurred seven weeks after admission.

POST-MORTEM EXAMINATION.—The relevant findings were as follows: *Lungs*: pink and well aerated, but emphysematous. In the *pleura* could be felt multiple

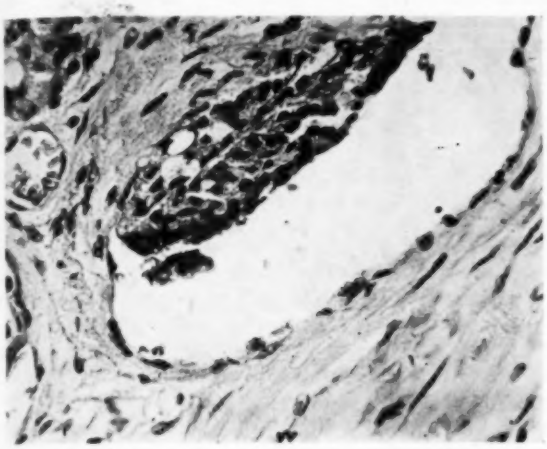

FIG. 303.—Tumour cells in a small venule. (× 250.)

small nodules and these were also scattered throughout the lung, mainly adjacent to bronchioles. *Alimentary tract*: normal as far as the ileocæcal valve. Here there was a tumour, 2·5 cm. in diameter, arising at the ileocæcal junction involving the upper lip of the ileocæcal valve and causing partial obstruction. The mucosa covering the growth was only superficially ulcerated and the mass was projecting into the lumen of the large bowel. On cutting, the tumour was moderately firm, homogeneous, and definitely yellowish in colour, with obvious infiltration of the muscularis, which was kinked but still showed a recognizable pattern. Immediately outside and continuous with it was what appeared to be a partly calcified neoplastic lymph-node 1 cm. in diameter (*Fig.* 302). Two small firm lymph-nodes were present along the ileocolic artery. The whole of the colon showed diverticulosis but no inflammatory masses. The *liver* was enlarged owing to multiple metastases, some of which were necrotic in their centres, and in two places confluent and forming large nodular masses in the right lobe lateral to the gall-bladder, and in the left lobe. These were not conspicuously yellow in colour. The morbid anatomical diagnosis was argentaffin carcinoma of the cæcum with multiple metastases.

HISTOLOGY.—1. *Ileocæcal tumour*: masses of cells were present infiltrating the whole thickness of the gut wall, causing the muscularis to be drawn inwards and kinked, with consequent protrusion into the lumen. The mucosa overlying the tumour was thinned, necrotic, ulcerated, and chronically inflamed, and showed numerous dilated capillaries containing fibrinous thrombi. Small masses of tumour cells were also present, and in some cases reached the surface of the mucosa. In parts the muscularis mucosæ was intact, though penetrated by tumour cells. The muscular layer was permeated by the growth, which in some places was replacing it and in others infiltrating along the planes of the muscle-fibres. The neoplasm had extended through the serosa into the adjacent tissues, in which was a partly calcified mass of fibrous and elastic tissue containing many large sclerotic arteries and veins and small lymph-nodes. Only one of the latter showed any deposit of growth, but several of the blood-vessels contained collections of neoplastic cells in their lumen (*Fig.* 303). The architecture of the tumour, in general, was fairly typical of argentaffin carcinoma. It was very cellular and had a well-formed vascular connective-tissue stroma which varied in amount in different parts. A noticeable feature was the intimate relation between the tumour and blood-vessels, and it is not surprising that hæmic dissemination took place.

Three main types of structure could be distinguished, viz.: (*a*) Masses of large regular cells with poor outlines

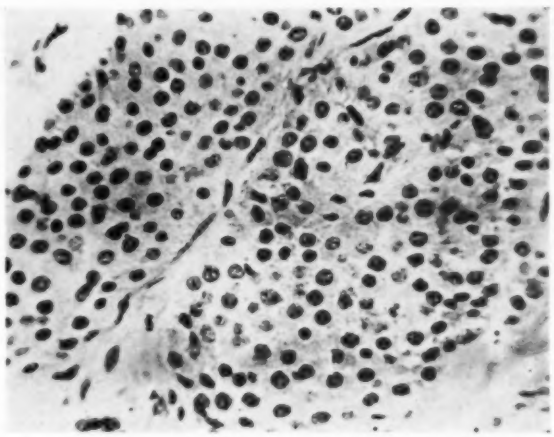

FIG. 304.—Type (*a*) structure. Undifferentiated neoplastic cells in large masses. (× 250.)

having finely granular cytoplasm staining pale pink with cosin, and large vesicular nuclei containing small punctate masses of chromatin. In some parts the cells were less regular and had hyperchromatic nuclei and occasionally were of bizarre nature or formed tumour giant cells. The connective-tissue stroma was rather delicate. Mitoses were not seen (*Fig.* 304). (*b*) Masses of smaller cells with

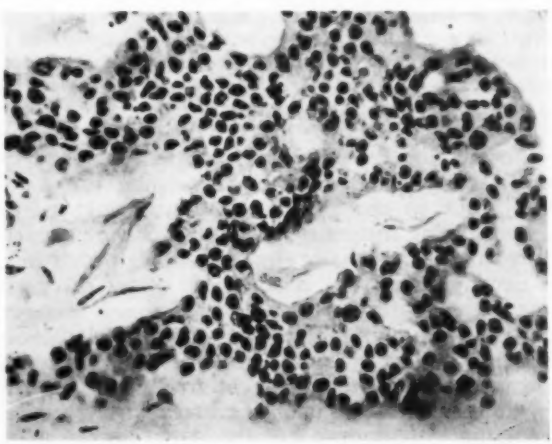

FIG. 305.—Type (*b*) structure. The rosette-like 'acini' are well shown. (× 250.)

granular but more deeply eosinophilic cytoplasm and deeper staining nuclei than (*a*). Occasionally, transitions between the two types of cell could be distinguished, but quite often they existed side by side, with a sharp dividing line. The nuclei showed much greater irregularity in morphology, but in places the cellular arrangement tended to be acinar, around a small central space containing granular eosinophilic material, the outer layer of cells being somewhat radially arranged. The stroma was well formed and included large bands of fibrous tissue (*Fig.* 305). (*c*) Masses of still smaller cells, with nuclei resembling those in type (*b*) but with less cytoplasm, which was not so eosinophilic. There was some acinar

arrangement, though in places this was lacking. Fibrous tissue was abundant, often cutting off the cells into small scattered islets. Spread along muscle planes was noticeable and transitions between types (*b*) and (*c*) could often be made out. In neither were mitoses found (*Fig.* 306).

2. *Metastases in liver and lungs*: The structure of these approached that of type (*a*) in the primary growth, but the tumour cells tended to be even less differentiated (*Figs.* 307, 308).

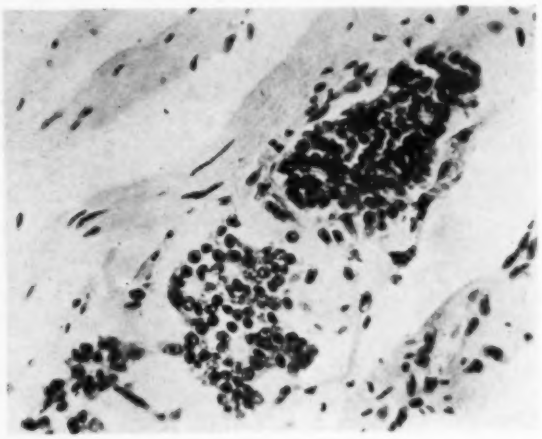

FIG. 306.—Type (*c*) structure. Tumour cells infiltrating the muscularis. (× 250.)

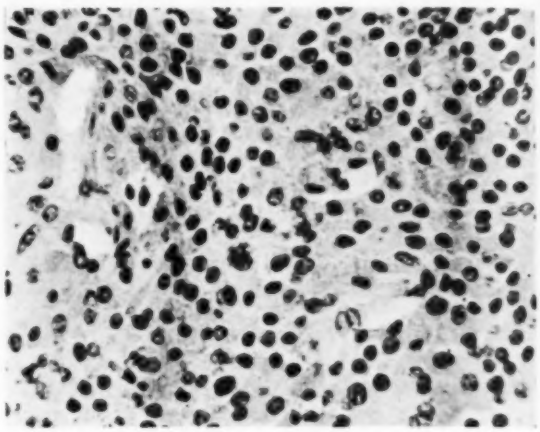

FIG. 307.—Metastasis in the liver. (× 250.)

HISTOCHEMICAL REACTIONS.—The primary tumour and the metastases were stained by Masson's modification of Fontana's silver method for argentaffin granules, and also by the diazo dye method of Clara as modified by Jacobson (1939). In none of the tissues could a satisfactory demonstration of argentaffin properties be made, but individual cells of types (*b*) and (*c*) gave definitely positive reactions with the diazonium salt and small groups gave a faintly positive reaction. The bulk of the neoplasm, however, showed no formation whatever of a diazo dye.

DISCUSSION

Obstruction is present in 50 per cent of the metastasizing ileal growths according to Cooke (1931), but in the present case was only partial, diarrhœa being the main symptom. Since ulceration of the mucosa is usually slight, tests for occult blood are usually negative. In the case under discussion these were positive on one occasion out of three. Judging by the amount of fibrosis and calcification, the tumour had probably been present for a long time and it is not surprising, in view of the patient's age, that metastasis had occurred. Metastasizing argentaffin carcinomas usually show spread to the regional lymph-nodes, and the liver is the next most frequent site of dissemination, but more distant organs are less often involved. Willis (1948) mentions lung and brain, and Cope and Newcomb (1930) reported a case with a secondary deposit in the epididymis. Ileal growths are responsible for the majority of widely disseminating argentaffinomas, and the first case to be recorded was of this type (Ransom, 1890). Although in the present case no help was obtained from radiography, a radiological sign has been described by Miller and Herrmann (1942). They state that a tumour at a point of kinking, with partial obstruction of the small bowel, is very suggestive of an argentaffin carcinoma.

Histologically, argentaffin carcinomas may show various degrees of differentiation, and also vary in their affinity for silver. It is generally recognized that these tumours arise from those cells of the intestine known by many eponyms (e.g., Nicolas, Kultschitsky, etc.) but actually discovered by Heidenhain (1870). Stout (1942) recommends the use of the name 'basigranular' cells as being the best term, since it avoids the use of proper names and does not presuppose any particular staining property. Their argentaffin capacity, first described by Gosset and Masson (1914), is not constant. Post-mortem changes reduce the likelihood of demonstrating the silver affinity, and after 8 to 10 hours after death it becomes increasingly difficult to elicit argentaffin properties. Jacobson (1939) states that the diazo dye reaction (Clara, 1934) is better than Masson's silver method for post-mortem material. This is in accordance with the findings in the present case, in which the autopsy was performed 24 hours after death. Some fixatives are better than others and not all silver preparations blacken the specific granules (Hamperl, 1932). The site of the tumour also influences the silver affinity.

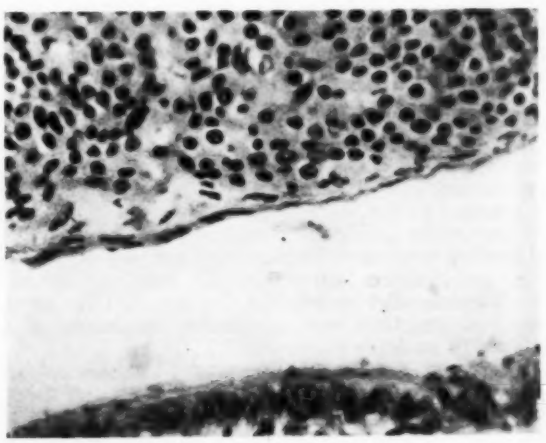

FIG. 308.—Metastasis in the lung, invading one side of a bronchiole and causing atrophy of the mucosa. The normal mucosa of the other side is seen in the lower half. (× 250.)

Of the 14 cases in the rectum reported up to 1949, only 2 gave a positive reaction according to Wilson (1949). He reported a further case in which only some of the tumour cells gave a positive result. Erspamer (1939) was of the opinion that the typical cells, which he called 'enterochromaffin' cells, are preceded by two earlier phases, in neither of which argentaffin properties can be demonstrated. Tumours of the latter will therefore not exhibit any affinity for silver. Such cases have been reported by Stout (1942) as carcinoids of pre-enterochrome cells. Pearse (1951) mentions an unpublished case of an argentaffin carcinoma of the stomach in which the specific granules gave the diazo dye reaction but did not reduce Fontana's silver solution in 24 hours. This conflicts with the findings of other authors that diazo positivity and argentaffin properties are related. It must be admitted, however, that histochemical studies have revealed a large number of different substances in these cells and there is no *a priori* reason why one or more should not be missing in some cases, with consequent effect on staining reactions. In the present author's case, the failure to exhibit silver affinity and to give a well-marked positive diazo dye reaction was almost certainly due to the long period elapsing between death and autopsy (24 hours). These reactions have been tried on other carcinoid material from ileum and appendix, and in each case positive results were obtained from tissues removed surgically (and therefore fixed with minimum delay) but not from the post-mortem material, the shortest time between death and autopsy being 16 hours.

SUMMARY

A case of argentaffin carcinoma of the ileocæcal region giving rise to metastases in the lungs and liver is described. A short discussion and review of the literature is given, with special reference to dissemination and histochemical behaviour.

I am grateful to Mr. Norman Matheson for the use of the case notes and to Mr. F. Goulden, A.R.I.C., of our Chemical Pathology Department, for making the Kernechtrotsalz B (4-nitro-*o*-anisidine diazotate) for the diazo dye reaction. I am much indebted to Professor W. D. Newcomb for criticism and advice and for the photograph of the specimen (*Fig.* 302), which was made in the Department of Medical Photography, St. Mary's Hospital, London. The photomicrographs were made by the author.

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MALIGNANT HEPATOMA

BY A. G. RIDDELL, LONDON

THIS paper records a case of malignant hepatoma with several unusual features and discusses the treatment of the case. The literature on the surgery of primary malignant tumours of the liver has been extensively described by Abel (1934) and Warvi (1945) so that a further review is unnecessary.

CASE REPORT

HISTORY.—A male hotel receptionist, aged 54, was admitted as an emergency to University College Hospital, complaining of epigastric pain of six hours' duration. After a gradual onset, the pain became continuous and severe, with periodic exacerbations, and radiated to the left costal margin and the tips of both shoulders. He vomited once, soon after the onset of the attack. There had been no disturbance of micturition and his bowels had been open normally on the day of admission.

He had no past history of abdominal pain, dyspepsia, or anorexia.

ON EXAMINATION.—He was pale and sweating; T. 96°, P. 100, R. 25, B.P. 118/80. There was generalized abdominal tenderness, most marked in the epigastrium, where there was some muscle guarding. There was no evidence of the presence of intraperitoneal fluid. Normal peristaltic sounds were heard on auscultation of the abdomen. Rectal examination revealed no abnormality.

The respiratory and cardiovascular systems were normal.

Investigations.—

Urinary Diastase—less than 2 units per ml.

Radiographic examination of chest and upper abdomen showed no evidence of pulmonary disease and no free gas under the diaphragm.

FIRST OPERATION.—

July 23, 1950: Exploratory laparotomy was carried out about eight hours after the onset of the patient's symptoms. On opening the abdomen through an upper right paramedian incision a considerable quantity of blood welled up into the wound. A white, friable nodule, about 1 cm. in diameter, which was bleeding, was seen in the free margin of the left lobe of the liver, and a second tumour of about the same size was palpable deep in the substance of the left lobe. There was no macroscopic evidence of cirrhosis and the remainder of the liver appeared normal. The tumour was excised from the free edge of the liver, bleeding being controlled with catgut mattress sutures, and the raw surface of the liver

covered with an omental graft. The wound was closed without drainage. Convalescence was uneventful. The tumour was examined microscopically and was reported as " a primary liver-cell hepatoma, which does not appear to be growing rapidly. The tumour is partially encapsulated but shows invasion of veins. The surrounding liver

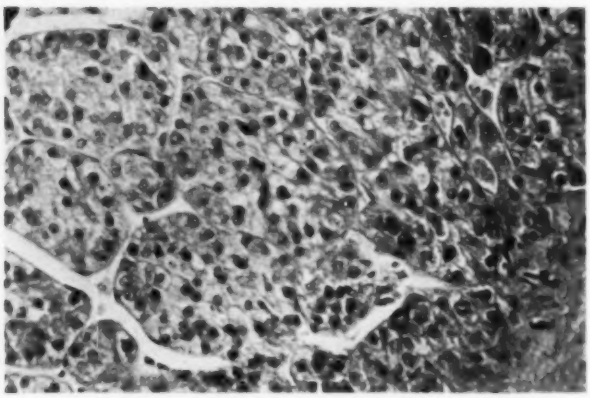

FIG. 309.—High-power view of primary tumour, showing hepatoma cells.

tissue shows cirrhosis and chronic hepatitis". (Section No. 1644/50.)

It was decided to attempt the excision of the left lobe of his liver.

SECOND OPERATION.—

Pre-operative Investigations.—

Serum-proteins 5·4 g. per cent. Hæmoglobin 88 per cent.

Empirical liver function tests—no evidence of liver damage.

Prothrombin concentration 95 per cent.

Blood-urea 30 mg. per cent. Urea clearance 70 per cent and 80 per cent.

Bromsulphthalein excretion test normal.

No excess urobilin(ogen) present in urine.

Lobectomy (Aug. 10, 1950).—Dissection lobectomy of left lobe of liver.

The abdomen was opened by a midline incision extending from the xiphisternum to a point halfway between it and the umbilicus; at the lower end of this incision a transverse incision was made outwards to the costal margin on each side. A few adhesions between the left lobe of the liver and the surrounding structures were divided. No other nodules were found in the liver and there were no secondary deposits on the peritoneum. The left triangular and the falciform ligaments were divided and the left lobe of the liver was then drawn up into the wound. The lesser omentum was divided and the porta hepatis exposed; no enlarged lymph-nodes were seen. The left branch of the portal vein, the left hepatic duct, and the left branch of the hepatic artery were isolated in turn and divided between silk ligatures. The lobe was then drawn downwards and the dissection continued . . e connective tissue between the liver and the inferior ven. . va until the hepatic vein from the left lobe in its extrahepatic course was found. It was about 0·5 cm. long, and was easily ligated and divided. Following ligation of the vessels the lobe became noticeably paler in colour. The liver was divided with the diathermy knife along a line from the porta hepatis to the left side of the inferior vena cava. The only bleeding came from a large vein in the posterior part of the liver, and this was easily controlled. The raw surface of the liver was covered with an omental graft. A drainage-tube was left down to the liver bed and the wound closed.

MORBID ANATOMY.—The left lobe of the liver was reported on as follows :—

" The left lobe of the liver weighs 215 g. In the centre of the lobe is a white nodule 1 cm. in diameter. Near the upper border of the liver is a second smaller nodule 2 mm. in diameter. There is an area of scarring and necrosis at the site of the previous operation. The remainder of the liver tissue appears macroscopically normal.

" Microscopic section of the two liver nodules shows a liver-cell hepatoma. The smaller nodule is an area of intravascular spread in a vein. Around both nodules there are veins and lymphatics containing tumour cells. (*Fig.* 310.) The surrounding liver tissue is normal in appearance." (Section No. 1785/50.)

PROGRESS.—There was no leakage of bile from the drainage-tube and it was removed on the second day. Apart from a partial collapse of the right lower lobe of his lung, convalescence was satisfactory. The patient was discharged on Aug. 23.

After discharge from hospital the patient remained well until December, 1951, when he complained of

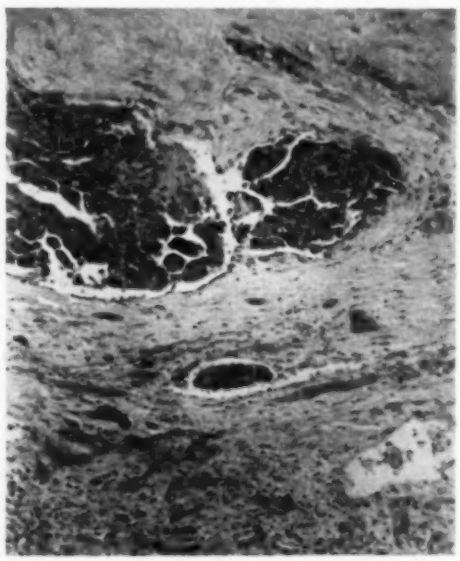

FIG. 310.—Secondary tumour, showing intravascular invasion.

abdominal discomfort. It was then found that his liver was considerably enlarged and that hard nodules were present on its surface. Although the patient is relatively well there can be little doubt that he now has a recurrence of his disease. Since his operation his liver function tests have been studied at regular intervals. These have shown a slight impairment of liver function, which has not increased.

COMMENT

Primary malignant tumours of the liver are rare in white-skinned races and are found in about 1 per cent of necropsies on cases of malignant disease (Willis, 1948). Liver-cell hepatoma is unusual, except in association with cirrhosis; if carcinoma of the liver occurs in the absence of cirrhosis it is more usually of the bile-duct type (Willis, 1948).

In this patient there was a localized area of cirrhosis and hepatitis around the nodule in the margin of the liver, but the rest of the liver was apparently normal on macroscopical examination, and this was confirmed on histological examination of the left lobe. It is probable that this area of cirrhosis was a local reaction to the tumour. It is unlikely

that there was a localized area of cirrhosis in an otherwise normal liver preceding the onset of the tumour.

Malignant hepatoma may be unicentric or multicentric in origin. In this case the relationship of the nodules to one another is uncertain. The nodule removed at the first operation and the other in the centre of the left lobe were about the same size and the character of size gives no indication as to which was the primary tumour. The third nodule, which was much smaller and was situated near the upper surface of the liver, was a secondary deposit in a vein. The palpable lump in the centre of the lobe was also probably a secondary deposit. There was some fibrous tissue around it that suggested the remnants of a vessel wall, but no smooth muscle cells could be identified. In the absence of a generalized predisposing disease of the liver it would therefore seem most likely that the tumour which bled was the primary growth and that the other two were blood-borne metastases.

It is rare for a hepatoma to rupture and cause bleeding into the peritoneal cavity. Specter and Chodoff (1950) reported 1 case due to a secondary tumour and collected 13 cases from the literature, of which all except 2 were primary liver tumours. On such occasions the symptoms produced are similar to those of a perforated peptic ulcer, acute cholecystitis, or acute appendicitis (Binkley, 1939). Acute symptoms can also appear when bleeding occurs into the substance of a hepatoma. One case has been observed in which it was not the primary tumour in the liver which bled into the peritoneal cavity, but a secondary tumour in the rectovesical pouch (Davies, 1950). The complication is usually fatal, only 3 out of the 14 reported cases survived the hæmorrhage ; only 7 of these cases were operated on (Specter and Chodoff, 1950). In this case the diagnosis on admission was uncertain. The absence of a previous history of dyspepsia and the diffuse abdominal tenderness without any marked muscular rigidity made the diagnosis of a perforated peptic ulcer seem unlikely. Acute pancreatitis was considered as a possible diagnosis, but was also unlikely as the urinary diastase was not raised. The physical signs were those of an intraperitoneal hæmorrhage except that the presence of intraperitoneal fluid could not be demonstrated.

The principal problems in the operative treatment of malignant hepatoma are the adequacy of the operation, the control of hæmorrhage, and the reduction in the amount of functioning liver tissue. Recent reports of left lobectomy are those of Pickrell and Clay (1944) and Freeman and Moreland (1948). In these cases the line of section was to the left of the falciform ligament and hæmorrhage was controlled by mattress sutures through the thickness of the liver. This type of operation has three disadvantages ; first, it is a removal of the anatomical rather than the morphological left lobe ; secondly, the liver tissue is difficult to suture and the sutures cause considerable tissue necrosis ; the third disadvantage of this method is that no attempt is made to remove the lymphatic glands from the hilum of the liver.

The method of lobectomy used in this case was that described by Raven (1949). It gives satisfactory control of bleeding, is a complete removal of the left lobe, and also facilitates the removal of lymphatic tissue from the hilum. The hilar dissection and ligation is straightforward and this alone would control the bleeding to a considerable degree. Studies in the post-mortem room have shown that sometimes the hepatic veins in their short extrahepatic course may be embedded in very dense connective tissue, and isolation of the left hepatic vein may be technically difficult. In such a case it should be abandoned and the resection of the lobe proceeded with and the bleeding radicles of the hepatic vein can then be individually controlled with encircling sutures.

An alternative approach to the liver is that of median sternotomy (Wangensteen, 1950). By means of this exposure much better access is afforded to the inferior vena cava and the hepatic veins and it is unnecessary to rotate the liver on its long axis. It was noted at operation that during the time that the liver was rotated on its axis and delivered into the abdominal wound, the patient's condition deteriorated, but rapid improvement occurred when it was replaced within the abdomen. This was probably due to traction on the inferior vena cava and impairment of the venous return. Care must be exercised to clamp the hepatic veins before dividing them to avoid air embolus (Wangensteen, 1950).

The possibility of precipitating liver failure by left lobectomy was considered pre-operatively. With a normal gross appearance of the liver and the inability to demonstrate impairment of liver function biochemically, it was decided that removal of the left lobe would not significantly affect the patient's liver functions. Further liver function tests carried out after the second operation did show some impairment of liver function. A comparison of the pre-operative and post-operative liver function tests have been reported by Flynn and Walshe (1951).

SUMMARY

1. A case of malignant hepatoma presenting as an acute abdominal emergency with a hæmoperitoneum is reported.

2. The technique of removal of the left lobe of the liver is discussed.

3. The tumour showed some uncommon pathological features.

I wish to express my indebtedness to Mr. H. R. I. Wolfe and Professor G. R. Cameron for their help in the management of the case and in the preparation of this paper.

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TRAUMATIC FEMORAL ARTERY THROMBOSIS

By G. E. MAVOR

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It is not generally recognized that calf claudication is the clinical manifestation of main vessel thrombosis. In the majority of cases the site of the thrombosis is in the femoral artery, while in the others the popliteal artery is thrombosed. Furthermore, where there is no nutritional upset of the limb at rest, and certainly no question of gangrene, it is almost invariably the case that where the femoral artery is thrombosed, the popliteal artery is patent, and where the popliteal is blocked, the femoral is free of thrombus. Only under these conditions of limited main vessel thrombosis does the collateral circulation, using all available channels, achieve an adequate by-pass function (Learmonth, Blackwood, and Richards, 1944). The main-stem artery is then refilled at a high level, thereby restoring circulatory normality as proximally as possible, and limiting the functional disability of the limb to periods of exercise. In such a limb, the degree of arterial insufficiency is determined by many factors: the initial extent of the thrombus; the degree of its subsequent propagation, i.e., the extent of the secondary thrombosis; and the collateral circulation. These factors are interdependent. Much depends on the collateral circulation and the speed with which it comes into action. It is formed initially from pre-existing channels, which are somewhat variable in size and location, and later it may be reinforced by new vessels, which are more numerous in younger patients. In most cases it follows a fairly distinct pattern. The main pre-existing channels are usually large, fairly constant, muscle vessels, now performing a new role; the later vessels are smaller and typically tortuous, often lying in close proximity to the main artery. Occasionally, however, the collateral circulation, as in the following case, may be unexpectedly interesting.

CASE REPORT

G. W., aged 33 years, a hairdresser by trade, was admitted to Aberdeen Royal Infirmary on Jan. 8, 1949. For two years he had experienced tightness in the right calf after walking. This complaint had become steadily worse, and on admission he stated that he could walk only about 100 yards at a slow pace without pain; thereafter he noticed tightness in the calf, relieved immediately by rest, but recurring after further exercise. To a less extent he was troubled by coldness of the right foot, especially after exercise, and on occasion he had noted that this foot was paler than the left. There was no rest pain and no history of trauma. While in the Army in 1942, he began to suffer from varicose veins of the right leg, which were cured by high ligation and several injections.

On Examination.—Significant changes were found in the right leg and foot. There was no abnormality of the skin at rest, but on exercise a patchy pallor of the right foot appeared, mainly of the first and second toes and adjacent plantar aspect. There was no muscle wasting and no diminution of power. The femoral pulse was strong, and equal to the left; there was no thrill or bruit. The popliteal and more distal pulses could not be palpated.

The Wassermann and Kahn tests were negative. The electrocardiogram showed no abnormality. The vasomotor reactions of the feet showed a normal response in the left great toe, and a very poor response in the right great toe, the skin temperature after seventy minutes rising only to 22·5° C. (*Fig.* 311). Femoral arteriography

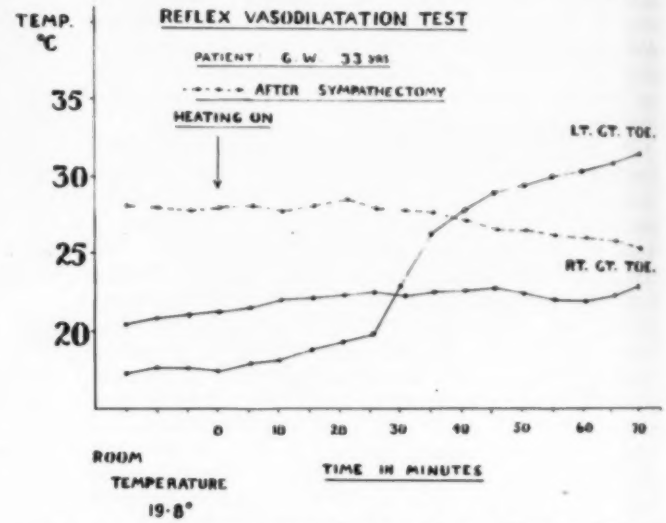

Fig. 311.—Vasomotor reactions of the feet after reflex vasodilatation, showing normal response of the left great toe. The vessels of the right great toe hardly dilate at all. After right lumbar ganglionectomy, the temperature of the right foot is maintained well above room temperature.

proximal to the profunda femoris origin, carried out under general anæsthesia, showed an extensive thrombosis of the artery stretching from the origin of the profunda to the point where the artery passed through the adductor magnus. Beyond this point the arterial tree appeared normal (*Figs.* 312, 313).

After viewing the arteriogram, the proximal 3 in. of the superficial femoral artery was resected in an endeavour to establish an accurate diagnosis. Notwithstanding its limited nature, the biopsy gives important information.

Histological Appearances (*Figs.* 314–316).—*Fig.* 314 shows the site of the initial thrombosis. Here the thrombus is in direct contact with the intima and organization has bridged the area between thrombus and intima. *Figs.* 315, 316 show sections from the area of the propagated clot, where the organized thrombus is completely free from the intima all around its circumference. No intimal lesion is seen. There is a variable amount of medial fibrosis. Thus the examination of the arterial segment shows that the thrombosis commenced just distal to the profunda origin, and that at this site there is no evidence of disease of the vessel wall.

Treatment and Progress.—An extraperitoneal right lumbar ganglionectomy, removing the second, third, and fourth lumbar ganglia and intervening chain, was carried out in order to aid the collateral circulation already well developed. Post-operative skin temperature recordings seven months after ganglionectomy showed that the temperature of the right foot was maintained considerably above environmental temperature, but, as expected, there was still no reflex vasodilatation (*Fig.* 311).

One year after ganglionectomy, the patient was satisfied with his condition. His right foot was always warm, his calf claudication was greatly relieved, and he could walk a mile to his work without difficulty except for slight pain localized to the lateral aspect of the calf. Apart from the increased skin temperature, there was no change on examination. A patchy exercise pallor of his foot persisted.

FIG. 312.—The arteriogram shows complete blockage of the femoral artery. There is great dilatation of the profunda femoris, which takes over the collateral circulation, the main connecting link being around the shaft of the femur via the last perforating artery and a muscle branch of the femoral. The popliteal is patent and well filled.

FIG. 313.—Filling in the lower leg is normal.

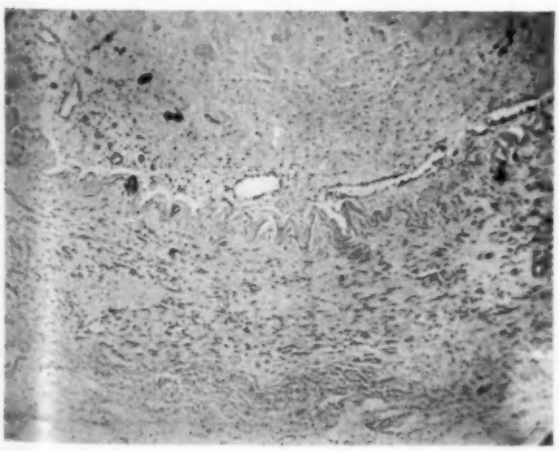

FIG. 314.—Section through the femoral artery, showing probable site of formation of the original thrombus, now undergoing organization. This is the upper end of the biopsy specimen. (× 67·5.)

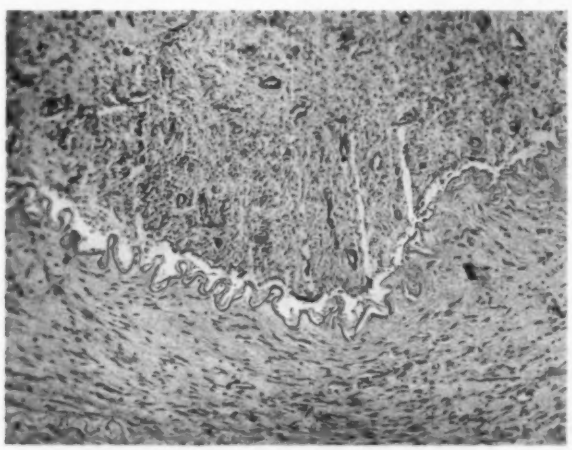

FIG. 315.—Section through the femoral artery at a lower level, showing the organization of the thrombus (or propagated clot) without attachment to the tunica intima. (× 67·5.)

Comment.—Although there is no history of trauma, nor of occupation or pastime causing a repeated strain on a certain part of the artery, biopsy findings prove that this is a case of traumatic femoral artery thrombosis. The patient's age and the unilateral lesion support this conclusion. In addition arteriography fails to demonstrate any evidence of disease or degeneration in the vessels visualized. In all probability the medial fibrosis visible followed the occlusion of the artery and the organization of the thrombus.

The most interesting feature of this case lies in the nature of the collateral circulation. It is extremely rare to see such a tremendous increase in the calibre and length of the profunda femoris. The whole profunda system has become greatly enlarged, and this enlargement includes many large muscle branches which strangely make no attempt to join in the collateral circulation.

There is little tortuosity and no short tortuous channels so often present in the younger age groups.

The collateral circulation fills up the main vessel at the opening in the adductor magnus, thereby fulfilling its by-pass function. It can be described in consequence as appearing adequate. This is supported on clinical grounds, as the resting needs of the limb are met apparently without difficulty. There are two points, however, which suggest that this surmise may not be altogether accurate: first, there is no peripheral vasodilatation in response to heating the trunk; secondly, the anterior and posterior tibial pulses are absent. While it is true that the popliteal pulse is absent in the presence of a main-vessel block, however short the thrombosis and however free the collateral circulation, the same does not hold for the more distal pulses; they on occasion may be palpable. Thus the arterial circulatory reserve of the limb is lower than it appears at first sight, related no doubt to the extreme narrowness of the vital collateral link to the femoral artery just beyond the distal extent of the thrombus.

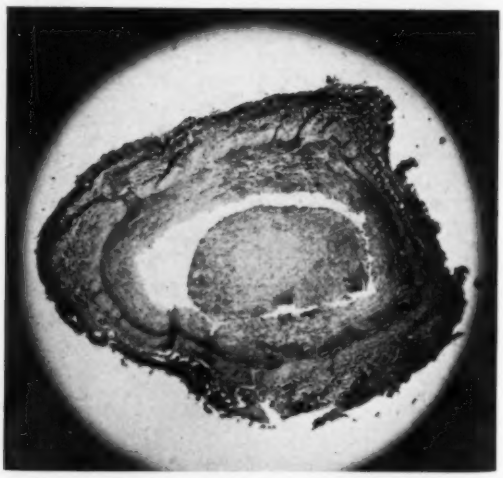

FIG. 316.—Section through the femoral artery at a lower level than *Fig.* 314, showing that the organized thrombus or propagated clot is completely free from the intima all around its circumference.

AETIOLOGY

Most cases of main-vessel thrombosis in the lower limb are secondary to atheroma. Youth in itself does not preclude such a diagnosis, for many cases of coronary artery thrombosis due to atheroma have been recorded in young men, and there is no reason to suppose that the femoral artery is a rare site for such pathological changes. Moreover, it is becoming increasingly apparent that atheroma may affect the arteries of the extremities of much younger patients than has hitherto been thought likely. The diagnosis in such cases is often regarded conclusively as thrombo-angiitis obliterans, and as a diagnosis this disease acquires a certain propriety in young people. Age alone can be no criterion, and in the presence of main-vessel thrombosis such a conclusion is likely to prove more speculative than accurate.

Other cases of thrombosis of the main artery are undoubtedly traumatic in origin, where there is direct damage to the artery, if not in continuity, certainly severe enough to initiate thrombosis. Dislocations, simple fractures, and wounds with or without fractures may be so complicated, a posterior dislocation of the knee-joint being probably the most common example of this peculiar type of injury. In other cases the trauma may be trivial or even obscure, apparently caused by minor sprains or habitual movements (Learmonth and others, 1944). Boyd (1949) considers that this explanation lies behind cases which he calls 'primary popliteal thrombosis', and of course the susceptibility of the popliteal artery to thrombosis is well known. In these cases the thrombosis is said to commence about the joint line level and to spread distally to the bifurcation of the artery or proximally to the adductor magnus opening (Boyd). Femoral artery thrombosis on the same basis of aetiology may not be as rare as Boyd seems to imagine. Here one has to consider muscle pull and compression as a factor, mainly in the region of the opening in the adductor magnus, a true muscle opening bounded by the hamstring and adductor portions of the adductor magnus, and not a tendinous arch or ring. At this point there is direct continuity of the general fascia of the muscles with the adventitia of the artery. In other cases the thrombosis commences just distal to the origin of the profunda femoris. This is not altogether a rare site for the onset of thrombosis secondary to atheroma of the intima, and it is possible that this part of the artery is exposed to some stress at present unknown.

It is important to consider these factors in detail as an accurate diagnosis should be reached wherever possible, if only to establish the prognosis. Obviously this is much more favourable in cases where trauma is a possibility than in those where the artery is diseased or degenerating. However, as these considerations of traumatic aetiology are in large part hypothetical, the diagnosis must remain in doubt in many cases. Learmonth groups such as cases of 'main-vessel thrombosis of indeterminate origin'.

Provided collateral vessels are preserved, biopsy is justifiable, since it may give a chance of establishing a definite diagnosis; but it may prove valueless, disclosing only a secondarily thrombosed vessel. If a long segment of the artery is thrombosed, biopsy should be carried out at the opening in the adductor magnus or just distal to the profunda origin.

I wish to thank Professor W. C. Wilson for advice in the preparation of this article. I am also indebted to Mr. G. Gordon Bruce for permission to publish the case; and to Mr. W. Carnie for the photography.

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EXOGASTRIC LEIOMYOMA

By J. HOWELL HUGHES and R. MARCUS, Liverpool

ALTHOUGH leiomyomata of the stomach are not rare, small tumours of this nature being not unusual incidental findings at autopsy (Golden and Stout, 1941; Meissner, 1944), cases presenting clinical symptoms due to this condition are comparatively uncommon. The report of 2 cases of exogastric leiomyomata is of interest as both tumours were large, extremely mobile, and, although both originated from the posterior wall, one was situated above, the other below, the stomach, and in both cases a long-standing atypical dyspepsia was relieved by removal of the tumours.

CASE REPORTS

Case 1.—HISTORY.—A married woman, aged 49, gave a history of vague dyspepsia of several years' duration. She complained of pain of a dragging nature situated under the right subcostal margin, worse at the end of the day, with some relief on lying down, and she had to release her corsets as the day progressed because of a feeling of fullness. During the last two years she had numerous radiographic investigations of the gastro-intestinal tract and gall-bladder which had proved quite normal.

ON EXAMINATION.—Her general condition was quite satisfactory and there was no evidence of loss of weight. On palpation of the abdomen a large tumour about the size of a fœtal head could be felt under the right subcostal region coming up from the loin. It was mobile and could be moved transversely in the pyloric plane to just beyond the midline and vertically as far as the umbilicus. When the patient was in the recumbent position the tumour disappeared under the subcostal margin and could not be felt again until it was pushed up from the loin. There was no tenderness and nothing else abnormal was found.

FIG. 317.—*Case* 1. The specimen. Painting of the tumour when delivered into the wound. The narrow pedicle containing the large blood-vessels is well demonstrated.

AT OPERATION (April 20, 1951).—On opening the abdomen through a right paramedian incision the gastro-colic omentum below the pyloric end of the stomach and the first part of the duodenum was seen to be raised by a mass situated in the region of the head of the pancreas. On palpation the mass was found to give a suggestion of lobulation, was extremely mobile, and was found to be in the lesser sac and extending under the right lobe of the liver. The omentum was freed from the transverse colon and the lesser sac opened. The tumour, dusky-red in colour (*Fig.* 317), was delivered into the wound and a pedicle ¾ in. wide was found connecting it to the posterior wall of the stomach just above the greater curvature and 1½ in. from the pylorus. The pedicle was clamped and divided at its attachment to the muscle coat of the stomach. The gastric mucosa was incised and exploration of the interior of the stomach did not reveal any other abnormality. The wound was closed with interrupted catgut sutures.

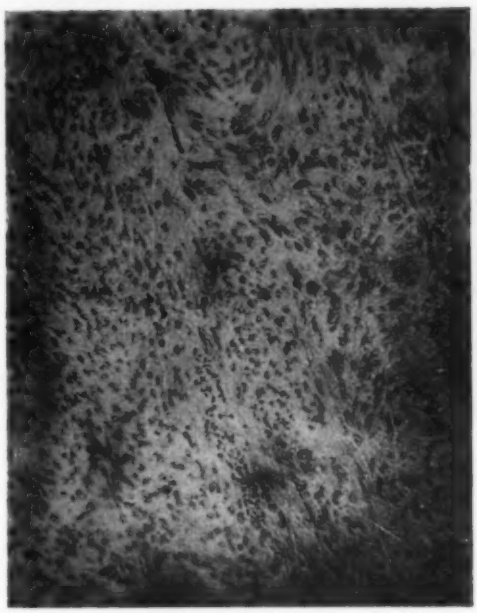

FIG. 318.—*Case* 1. Photomicrograph of a section of the tumour, showing typical leiomyomatous tissue. (× 136.)

POST-OPERATIVE PROGRESS.—The convalescence was uneventful until the eleventh day, when a mild infection in the lesser sac occurred. This resolved completely after a five-day course of streptomycin, and the patient was discharged on the twenty-sixth post-operative day, her condition being quite satisfactory.

PATHOLOGICAL REPORT.—The tumour weighed 425 g. and histologically was a leiomyoma (*Fig.* 318).

Case 2.—A male patient, aged 70 years, presented with a history of flatulence, fullness after food, and slight loss of weight over a period of two years. He had no loss of appetite and no actual abdominal pain, but had been diagnosed elsewhere as an inoperable carcinoma of the stomach.

ON EXAMINATION.—The signs of loss of weight were only slight and he looked fairly fit for a man of his age. His tongue was clean, and on palpation of the abdomen a large hard mass with a regular outline could be felt in the epigastrium. The mass was about the size of a fœtal head, was freely mobile, and could be pushed from just below the left subcostal margin to a similar position on the right side of the abdomen. During the examination it occasionally slipped below the left lower ribs and was no longer palpable.

Radiological Examination was carried out by Dr. P. H. Whitaker on Jan. 14, 1952, who reported: "There is a rounded defect in the lower third of the body of the stomach. The margins are clear-cut. The mass is mobile and alters its relationship to the outline of the stomach although it maintains a constant relationship to

the fundus. The appearances would be consistent with an extrinsic mass closely associated with the stomach, such as a pedunculated subperitoneal gastric tumour." (*Figs.* 319, 320.)

FIG. 319.—*Case* 2. Radiograph taken in prone position immediately after opaque meal, showing normal outline of rugæ of the stomach.

AT OPERATION (Jan. 15).—Midline incision. A large nodular lump varying from a deep red to plum colour was seen stretching the lesser omentum and deforming

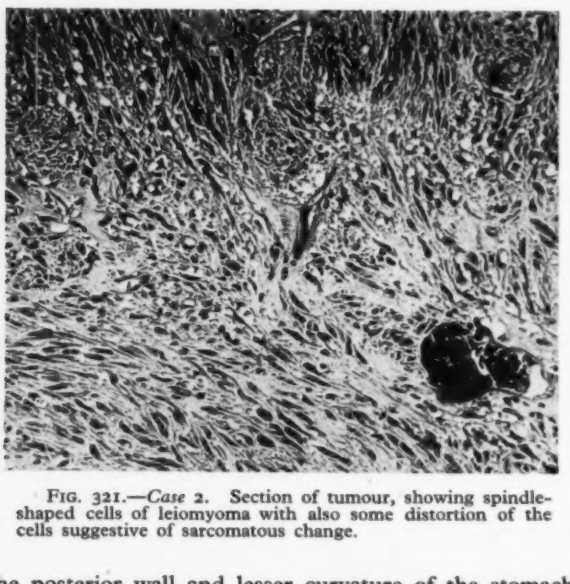

FIG. 321.—*Case* 2. Section of tumour, showing spindle-shaped cells of leiomyoma with also some distortion of the cells suggestive of sarcomatous change.

the posterior wall and lesser curvature of the stomach. The lesser omentum was opened and the tumour brought into the wound. The tumour was attached by a pedicle about ¾ in. wide to the posterior wall and there were large vessels running in the pedicle. These were divided and the portion of the whole of the stomach wall forming the base of the pedicle was excised and the stomach wall closed in layers.

The patient made an uninterrupted recovery and was discharged on the fourteenth post-operative day.

PATHOLOGICAL REPORT.—The tumour weighed 500 g., and measured 14 × 11 × 10 cm., and histologically was a leiomyoma undergoing doubtful sarcomatous change (*Fig.* 321).

FIG. 320.—*Case* 2. Radiograph taken in erect position immediately after opaque meal, showing rounded filling defect of lesser curvature.

DISCUSSION

Leiomyomata arise from the muscular coat of the stomach and may be exogastric or endogastric, depending on their position relative to the lumen of the stomach (Golden and Stout, 1941), and a characteristic feature of these tumours is their mobility. In the endogastric type which grows towards the lumen, this mobility may lead to partial invagination of the stomach wall, ball-valve obstruction of the pylorus (Willis, 1948), or a gastroduodenal intussusception (Elgood, 1951). In this type the overlying mucosa, and sometimes the tumour, undergo ulceration and as the result of insidious bleeding severe anæmia, sometimes mistaken for pernicious anæmia, can occur. In practically all the described cases in which hæmorrhage was a dominant feature, ulceration was present in the fundus of the tumour (Sworn and Cooper, 1938; Whyte, 1940; Levy and Horn, 1941), and the radiological findings in such cases are said to be sufficiently characteristic to make diagnosis possible. In one series over 50 per cent of the cases presented with hæmatemesis or melæna (Golden and Stout, 1941). Dyspepsia of indefinite character is another common feature and in a series of 39 cases of gastric leiomyomata it occurred in 8 cases (Behrend, 1944). As expected, the free and

total acidity of the stomach is normal and this, combined with the long history and the comparatively slight alteration in the patient's weight, renders it possible to differentiate the condition from malignant disease of the stomach, even when the tumour produces a gross filling defect of the stomach on radiological examination.

The exogastric type grows away from the lumen of the stomach, becomes subserous, and then grows into the peritoneal cavity. Willis (1948) recorded a subserous gastric myoma weighing 900 g. and Rajasingham and Cooray (1950) successfully removed an unusually large leiomyoma of the stomach which nearly filled the abdominal cavity and weighed 4080 g. This type may give rise to atypical dyspepsia with negative radiological findings and it depends on the size of the tumour whether a lump is palpable on abdominal examination. When found it is usually mobile, non-tender and diagnosis is only certain on exploration of the abdomen.

SUMMARY

Two cases of gastric leiomyomata presenting in the lesser sac and removed at operation are reported. The clinical features of the two types of leiomyomata of the stomach are recorded.

Our thanks are due to Dr. P. H. Whitaker for the radiological report and prints; Miss Barbara Duckworth, Artist to the Department of Surgery, Liverpool University, for the painting; and Mr. Wilfred Lee, of the Central Photographic Department, Liverpool University, for the photomicrographs.

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ACCESSORY SPLEEN IN THE SCROTUM

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OUT of 12 previously reported cases of accessory spleen in the scrotum, only the first 2 are to be found in the British medical literature (Allbutt and Rolleston, 1908; Sneath, 1913). According to Putscher (1934), Bostroem in 1883 demonstrated a growth joining the spleen and the left testicle with both organs in their normal position, but Putscher was unable to find a record of the case. The first definite case was found at post-mortem by Dr. A. Latham at St. George's Hospital and recorded by Rolleston, who was shown a long process of splenic tissue bound down to the posterior abdominal wall and passing into the left side of the scrotum to the testicle. Sneath (1913) was unaware of any previous case and his was regarded as the first record of this anomaly by subsequent American and other foreign writers (*Fig.* 322). Pommer (1888) described and illustrated a band of accessory spleen passing from the splenic hilum to an undescended testicle situated in the left iliac fossa; this, however, was noted at the post-mortem of a baby without legs and associated with other gross congenital deformities. Skworzoff (1924) found a similar condition in a boy aged 10 with congenital deformities of arms and legs; the left testicle in this case was in the groin. Females are not immune from the splenic abnormality; Wiltschke (1929) found in a newborn girl a splenic cord 77 mm. long which passed from the upper part of the hilum of the spleen to the mesovarium.

CASE REPORT

A boy, aged 7, was first seen in 1946 when he was referred to an out-patient clinic on account of a tender enlarged left testicle; the right testicle was thought to be undescended.

The boy's mother reported history of pain in the left testicle for about one month. She thought the left testicle had been swollen from birth, but she was rather indefinite on this point.

The right testicle was found to be normal but retractile. The left testicle was definitely enlarged and the left spermatic cord was thickened and irregular. Rectal examination was negative and his urine was clear of infection. A diagnosis of a partial torsion or thrombosis of the spermatic cord was made. He failed to re-attend for further observation as instructed.

In 1949 he returned owing to recurrence of left inguinal pain aggravated by exercise. Our colleague, Mr. Brewer, having found the condition of the left testicle and cord unchanged, kindly referred him back for review. A full urinary investigation appeared necessary in view of the possibility of tuberculosis. An intravenous pyelogram and examination of a 24-hour specimen of urine, however, showed no abnormality. Exploration was later considered advisable owing to continued pain, and a diagnosis of diffuse lymphangioma of the cord was suggested.

AT OPERATION.—On Dec. 18, 1950, the left spermatic cord was exposed through an inguinal incision. A thin, narrow, true congenital hernial sac was present, with normal spermatic vessels and vas deferens. Inside the sac was a purple cord fused to the testicle below. The upper end of the cord, which had a variable diameter, was found to be lying free in the neck of the sac. It was then discovered that on traction of the upper end of the cord a large respiratory excursion occurred suggesting an attachment to the diaphragm. The abdomen was opened in the midline and the cord was traced from the internal abdominal ring through the peritoneal cavity, without a mesentery, to the region of the hilum of a normal-looking spleen. The fusiform termination of the cord, which had the same colour as the spleen, was ligated and divided. The spermatic vessels and vas deferens were divided before removal of the left testicle,

congenital hernial sac, and cord in one mass. Before closure of both incisions it was noted that both kidneys felt normal.

His convalescence was uneventful apart from two days' unexplained left abdominal pain two months later. He is now well.

THE SPECIMEN.—After fixation in formol saline the specimen measured about 8 in. in length, probably being somewhat longer when unfixed (*Fig.* 323). At its upper end it was ½ in. in width. In the middle one-third of its length it narrowed to about ¼ in., expanding, as it reached the testis, to about ½ in. The testis itself was about ¾ in. long and there was little macroscopic line of demarcation between it and the column of abnormal tissue. The latter was covered by a thin glistening capsule.

FIG. 322.—Sketch of the spleen, accessory spleen, and left testicle. (*Sneath's case re-drawn.*)

Histological examination revealed that this reddish cord was splenic tissue. All the elements of normal spleen were present. In the upper and lower ends the architecture appeared to be normal. In the middle third there was a considerable excess of fibrous-tissue stroma.

At its lower end the splenic tissue abutted on to the body and mediastinum of the testis. A well-defined fibrous band divided splenic tissue from the main mass of testicular tissue (*Fig.* 324). In one place, however, a group of seminiferous tubules lay on the splenic side of the band and were immediately surrounded by splenic lymphoid tissue (*Fig.* 325). The fibrous band between spleen and testis was continuous with the capsule over the rest of the cord of splenic tissue and appeared to be normal splenic capsule. It tended to merge with the testicular connective tissue, but the tunica albuginea was missing in this region although present over the rest of the testis. The seminiferous tubules appeared normal for the patient's age but the rete testis and efferent ducts were distorted and compressed.

FIG. 323.—Complete cord of accessory spleen fused to the left testicle lying in a congenital hernial sac.

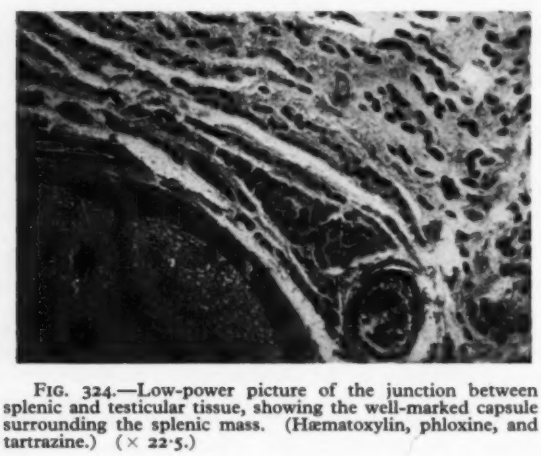

FIG. 324.—Low-power picture of the junction between splenic and testicular tissue, showing the well-marked capsule surrounding the splenic mass. (Hæmatoxylin, phloxine, and tartrazine.) (× 22·5.)

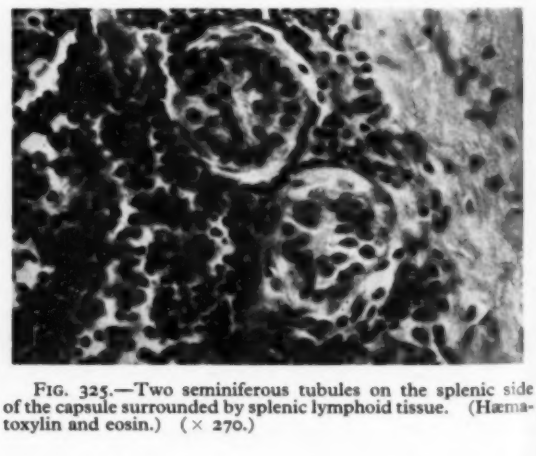

FIG. 325.—Two seminiferous tubules on the splenic side of the capsule surrounded by splenic lymphoid tissue. (Hæmatoxylin and eosin.) (× 270.)

DISCUSSION

Accessory spleens have been long recognized by pathologists, who report their frequency in post-mortems as being at least 10 per cent (Adami and Nicholls, 1911; Helwig, 1929), while an incidence of 35 per cent has been stated (Morrison, Lederer, and Fradkin, 1928). The majority of accessory spleens occur in the region of the pedicle of the normal spleen, but they have been found in the greater and lesser omenta and embedded in the tail of the pancreas (Robertson, 1938). Occasionally many tiny splenules have been found in the peritoneum, mesocolon, and pouch of Douglas.

Table I.—Cases of Accessory Spleen in the Scrotum

Author	Year	Age	Mode of Discovery	Type of Cord	Presence of Hernial Sac	Comments
Bostroem	1883	?	Post mortem	Complete	Probably yes	
Allbutt and Rolleston	1908	?	Post mortem	Complete, but retroperitoneal	Yes	
Sneath	1913	Adult negro	Post mortem	Complete intraperitoneal, partially fibrous	Yes	
Heitzmann	1917	25	Operation for left inguinal hernia	Complete, intraperitoneal	Yes	
Finaly	1926	Baby	Operation for left inguinal hernia	Absent	Yes	
Talmann	1926	22	Operation for epididymitis	Absent	No	One nodule attached to epididymis and another splenule in spermatic cord
Osselladore	1929	19	Operation for ? third testicle	Absent	No	Three splenic nodules which enlarged during malarial attacks
Fischer and Gissel	1935	13	Operation for left inguinal hernia	Traced to internal ring	Yes	
Settle	1940	Adult	Operation for epididymitis	Absent	No	The left testicle enlarged during malarial attacks
Emmett and Dreyfuss	1943	47	Operation for ? tumour of left testicle	Absent	Yes, bilateral	
Olken	1945	56	Post mortem. Hæmatemesis from duodenal ulcer	Complete, intraperitoneal	Yes	Another splenule present near splenic hilum
Andrews and Etter	1946	32	Operation for ? epididymitis	Traced to external ring	Yes	
Tate and Goforth	1949	21	Operation for ? tumour of left testicle	Traced to external ring	Yes	
Bennett-Jones	1952	10	Operation for ? lymph-angioma of cord	Complete intraperitoneal	Yes	

Aetiology.—In the lower animals the splenic formation is not confined to the dorsal mesogastrium; the spleen spreads caudally even into the mesentery of the hind gut (Keith, 1933). This could explain the occurrence of an accessory spleen in the pelvis, but there is some evidence (Hambrick and Bush, 1942; Krueger and Mast, 1942) that an injury to the spleen might be followed by multiple splenic autografts in the peritoneal cavity; if this is the mode of production, there ought to be definite signs of such an injury to the normal spleen. No injury could explain the occurrence of splenic tissue attached to the testicle. In four of the recorded cases there was apparently no cord passing between the splenic tissue in the scrotum and the normal spleen, but in one case another separate splenic nodule was present in the spermatic cord (Talmann, 1926). A complete band from a normally placed spleen and left testicle has been observed six times, including Bostroem's case and our own. From the descriptions and illustrations these bands were very similar (*Fig.* 326); the diameter varies, but the girth is larger at both extremities while the middle portion is narrow and actually fibrous in one case. All the complete bands,

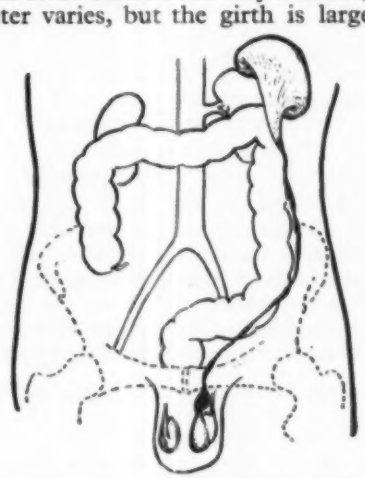

Fig. 326.—Sketch showing the relationship of the accessory spleen to the abdominal viscera. (*Olken's case re-drawn.*)

with the exception of Allbutt and Rolleston's case, passed through the peritoneal cavity without a mesentery to the spleen. The exact mode of attachment to the spleen varied ; some were attached to the anterior border, while others were attached to the hilum. A true congenital hernial sac appeared to be present in all those cases with complete bands, but its presence

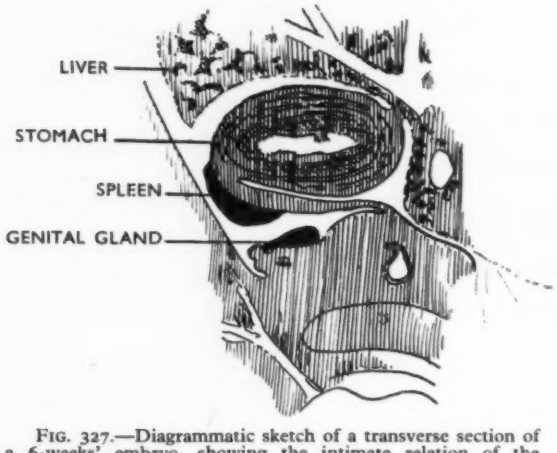

FIG. 327.—Diagrammatic sketch of a transverse section of a 6-weeks' embryo, showing the intimate relation of the developing spleen to the genital gland. (*After Sneath.*)

was not always specifically mentioned. An explanation of the course of the band through the peritoneal cavity without a mesentery has been suggested by Sneath. In a six-weeks-old embryo the rudimentary spleen in the dorsal mesogastrium lies opposed to the genital gland lying upon the posterior abdominal wall (*Fig.* 327). He presumed that any slight inflammation of the peritoneum occurring at this stage might cause an adhesion between the spleen and the genital gland.

Clinical Significance.—Accessory spleens in the abdomen have been known to undergo torsion, giving rise to acute abdominal pain (Alexander and Romanes, 1914 ; Alexander, 1929) or even intestinal obstruction (Voss, 1934). They may add to the difficulty in ligation of the splenic pedicle during splenectomy or isolation of the splenic vein for a lienorenal anastomosis. Failure to remove accessory spleens has been held responsible for failure to cure thrombocytopenic purpura in 15 per cent of cases (Morrison, Lederer, and Fradkin, 1928) by splenectomy. In acholuric jaundice accessory spleens may also prevent a cure by splenectomy.

In the scrotum an accessory spleen has been mistaken for an inguinal hernia containing omentum, a third testicle, a testicular tumour, torsion of the testicle, thrombosis of the spermatic vessels, tuberculous and non-specific epididymo-orchitis, and a diffuse lymphangioma of the spermatic cord. There has been no correct pre-operative diagnosis even in the two cases where the testicular swelling enlarged during malarial attacks (Talmann, 1926 ; Settle, 1940)!

SUMMARY

Fourteen cases of accessory spleen in the scrotum have been collected. The four autopsy cases and two operation cases were found to have complete bands of splenic tissue between the normal spleen and the left testicle. In the remaining eight operation cases a mass of splenic tissue was removed from the left side of the scrotum and in three of them a band was traced upwards only to the internal or external abdominal ring ; it is likely that these bands passed to the spleen. Our case is apparently the first to have a complete accessory spleen attached to the spleen and a normally placed left testicle removed during life. Heitzmann (1917) was the first to trace the cord upwards into the abdomen during an operation.

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A CASE OF TRUE HERMAPHRODITISM

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CASE REPORT

THE patient, a Hindu of 14 years of age, was sent from India with a diagnosis of hypospadias. In May and June, 1950, a successful two-stage operation was performed, but the general physical conformation of the patient having aroused suspicion he was referred to one of us (R. G.) in July, 1950.

He is the youngest of four children, the elder ones being normal girls. No change had occurred in the health of the mother which might lend credence to the view that her hormonal pattern had prejudiced the normal development of this individual child. She has since given birth to a normal son. He had always passed urine from the under side of the proximal end of the penis, but, apart from this, no abnormality had been suspected until about a year before, when the development of his breasts had begun to cause him much embarrassment. He had suffered from no serious illness, but had lost the sight of his right eye through accidental damage to the cornea. He had never overtly menstruated.

FIG. 328.—General appearance, showing feminine distribution of fat and pubic hair, female breasts, small penis, and testicle in the right side of the scrotum.

He was extremely obese, the distribution of the adipose tissue being somewhat feminine (*Fig.* 328). He had well-developed female breasts and, as one would expect at his age, a feminine distribution of pubic hair and very little body hair. The scrotum contained one gonad, on the right side. There was no vagina, but a uterus could be felt by bimanual examination. The phallus was about 2 in. in length, and, as a result of operation, the urethra, formerly hypospadiac, now opened at the tip (*Fig.* 329). Psychologically, he was entirely masculine in his outlook. Unlike many boys of his age and race, he was sexually 'unawakened', but had the normal outlook of a European boy of his age towards girls, and enjoyed boyish pastimes. He foresaw himself as the father of a family.

It was clearly necessary to establish with certainty his true sex, and on July 21, laparotomy was performed by one of us (D. M.). A lower right paramedian incision was made. The layer of subcutaneous fat was approximately 4 in. thick. Pelvic examination revealed an ovary on the left side of normal adult size and appearance. The left Fallopian tube and fimbria were also of normal appearance. There was a small uterus of approximately normal shape. It was deflected towards the ovary. There was no trace of the right ovary or tube except for a tiny thin cord which ran outwards from the uterus for about an inch.

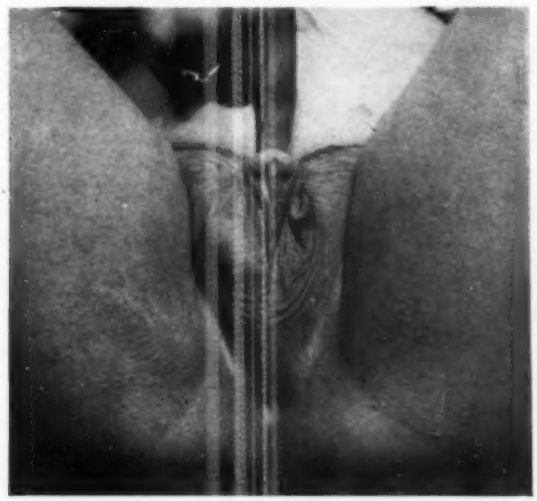

FIG. 329.—Showing original hypospadiac condition and empty scrotum on the left side.

The uterus was removed together with the left ovary and tube. There was no vagina, but a thin solid cord of tissue descended from its lower margin through the cellular tissues towards the back of the neck of the bladder. It ceased to be recognizable before it reached the bladder or urethra.

A biopsy was then performed on the gonad in the right side of the scrotum. This appeared to consist of normal testicular tissue. The epididymis and the lower

end of the vas were inspected and palpated through the scrotal incision. Both were normal.

Convalescence was uneventful and a bilateral, simple mastectomy was performed on Aug. 22, 1950. The nipples were grafted as free grafts after trimming away the excess skin. Both took satisfactorily and he was discharged on Sept. 5. Both breasts consisted of fat and breast tissue in normal amounts.

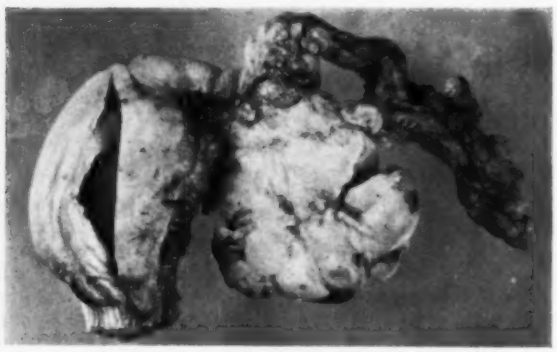

FIG. 330.—Unicorn corpus uteri and left adnexæ, anterior view.

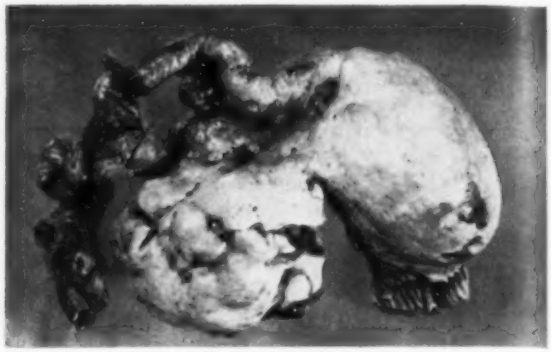

FIG. 331.—The same, posterior view.

PATHOLOGICAL FINDINGS.—

The *specimen*, received fresh, consisted of a subtotal unicorn uterus with adnexæ (*Figs.* 330, 331) and a small dice of testis. The testis, a piece of endometrium, and two pieces of ovary were fixed in 25 per cent v./v. glacial acetic acid in absolute alcohol, the rest of the material in 10 per cent formol saline. After fixation the uterus measured 4·5 × 2·5 × 2 cm., the tube 7·5 cm. in length, the ovary 3·5 × 3 × 1·5 cm.

The cavity of the uterus was slightly dilated and contained fresh bloody fluid: it appeared to end blindly below, though the crushed lower pole made this difficult to decide. The muscular wall was 5–8 mm. thick, the mucosa smooth and 0·5–1·0 mm. thick. The adnexæ were essentially normal. The ovary showed a blood spot in the middle of its antihilar border: the rest of the surface had the usual smoothly bosselated appearance. Its cut surface was everywhere unremarkable and showed nothing suggesting testicular tissue.

Microscopically, the endometrium was of late menstrual (*c.* third-day) type. The glands were of involuted secretory form (*Fig.* 332), their cells showing basal nuclei and pale washed-out cytoplasm. The stroma was dense, with a mild superficial leucocytic infiltration: it contained no pigmented or ferrocyanide-positive cells. The surface was partly epithelialized, partly bare, with fragments flaking off: remains of spiral arterioles could be seen just beneath it (*Fig.* 333). At the lower pole the mucosa was similar, and serials at what appeared to be its lowest point showed no extension into the clamped region and no isthmic or cervical mucosa. Both here and above, the rest of the wall was purely muscular.

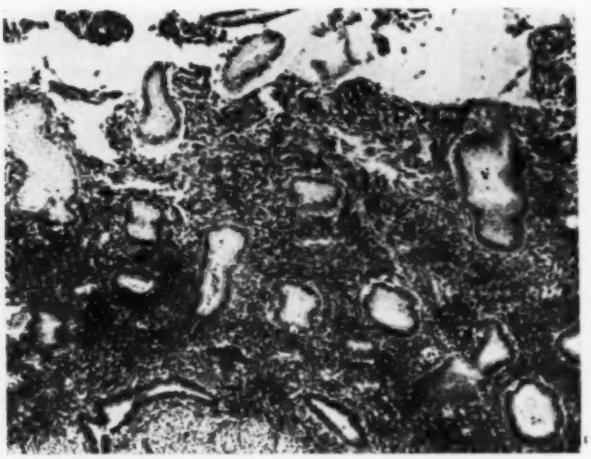

FIG. 332.—Late menstrual endometrium, showing involuted secretory glands, dense stroma, myometrium below, and flaking surface above. H. and E. (× 75.)

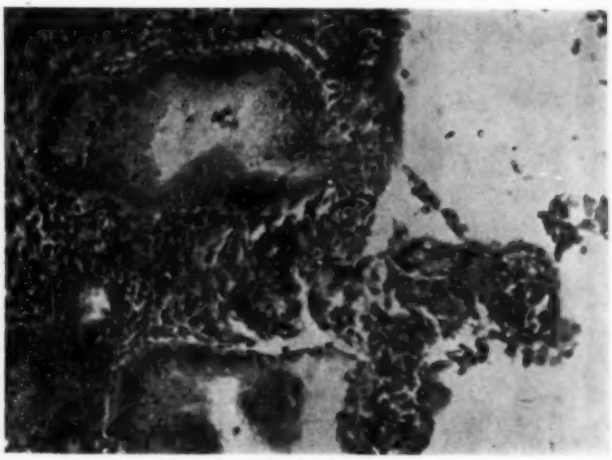

FIG. 333.—The same, top right, at higher magnification, showing gland cell detail, leucocytic infiltration, partly epithelialized surface and head of spiral arteriole. (× 180.)

The isthmic (*Fig.* 334) and ampullary (*Fig.* 335) parts of the tube appeared normal. The former was lined by tall columnar epithelium; the latter partly by tall, but mainly by low, epithelium appropriate to the phase of the cycle. The lumen contained a few red cells, polymorphs, and cellular detritus.

A single epoöphoron canal, located with difficulty, appeared to be normal. A few epithelialized peritoneal inclusions, surrounded by effused red cells, were seen in the mesovarium.

Blocks were prepared from the ovary from across the centre, from the stigma, and from the medial and lateral parts of the hilus: the last three of these were cut serially. In the lateral block a single nest of ovarian hilus (paraganglionic, Leydig) cells was found (*Fig.* 336) in the mesovarium; at the hilus were normal rete and 'medullary canals' (*Fig.* 337). The last had the form of a series of vesicles arranged roughly in a line parallel

to the tube and perpendicular to the plane of section. They were partly separate, partly linked up as in *Fig.* 337, were of smooth contour, and lined by dark cubical epithelium. An occasional follicle at the stage of a single epithelial granulosal ring was found in line in this series. No quasi-testicular tissue was seen in this or any other block.

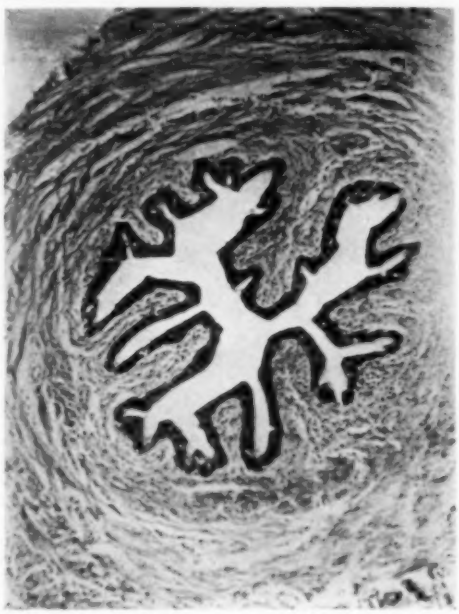

FIG. 334.—Fallopian tube, normal isthmus. H. and E. (× 67·5.)

FIG. 335.—Fallopian tube, part of normal ampulla, showing both low and tall epithelium. H. and E. (× 86·25.)

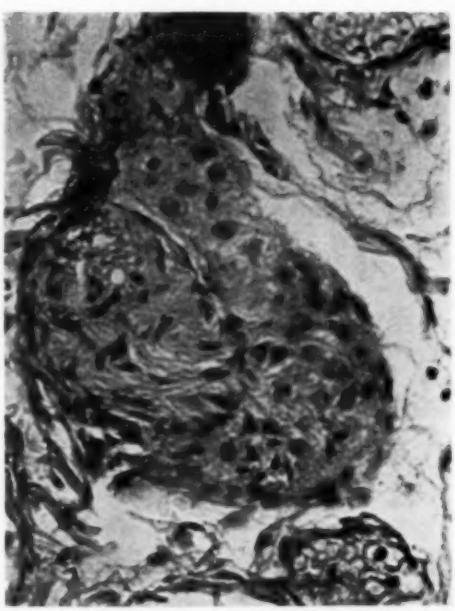

FIG. 336.—Small nerve-fibre in mesovarium with crescentic cuff of ovarian hilus (paraganglionic) cells. H. and E. (× 270.)

FIG. 337.—Hilus of ovary showing rete below, remains of follicle in late atresia above, and medullary canals just below this. H. and E. (× 21.)

The general architecture of the ovary (*Fig.* 338) appeared normal, but with more than the average number of late ripening follicles. Primordial, maturing (*Fig.* 339), and atretic follicles were readily found, as well as corpora fibrosa, but no corpora albicantia. The stigma showed an ovulation plug (*Fig.* 340). Beneath it lay a corpus luteum in an early stage of involution. It was of roughly normal structure but less folded, with smaller granulosa-lutein cells (*Fig.* 341) and more connective-tissue fibres (*Fig.* 342) than the average.

The testis showed some tubules (about a fifth) in all stages of atrophy up to complete hyalinization, but the majority resembled those of a healthy young adult (*Fig.* 343). All phases of the germ-cell lineage up to spermatids and spermatozoa were seen (*Fig.* 345). In many tubules nearly all the spermatogonia and spermatocytes were in some phase of cell division (*Figs.* 344, 345). The number of interstitial cells seemed above the average but not pathologically so: in these there were only occasional mitoses (*Fig.* 344). No Reinke crystalloids were seen.

The breasts were grossly and microscopically similar to those of a normal female in architecture (*Fig.* 346), but showed some periductal lymphocytic and plasma-cell infiltration.

Chromosome Constitution of the Testis.—The endometrium showed no cell divisions, but portions of acetic-alcohol-fixed testis and ovary were used by one of us (A. H.) for cytological study.

Feulgen squash preparations were made of the testis material (12 minutes' hydrolysis in *N* HCl at 60°). The

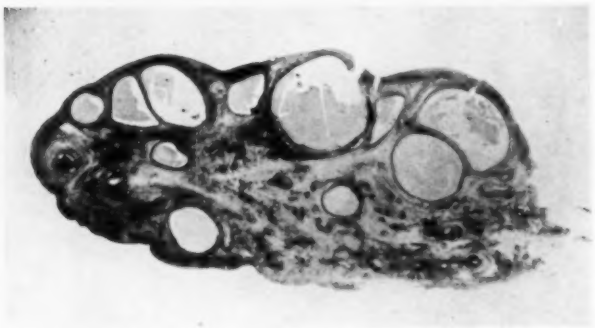

FIG. 338.—Ovary, transverse section, showing general architecture. H. and E. (× 2·2.)

tissue was somewhat alcohol-hardened and considerable difficulty was encountered in finding cells which were adequately spread out. Eleven first meiotic metaphases form the basis for the following remarks on the chromosome constitution of this tissue. The number of bivalents is apparently 24; in no cell could a completely unambiguous count be made, but there is no reason for believing that the chromosome count differs

FIG. 340.—Section of stigma showing cavity of corpus luteum to right, scroll of fibrin in centre, effused blood to left, edge of granulosa-lutein layer at bottom right. H. and E. (× 30.)

from normal. Some first meiotic metaphase cells show two unequal elements lying off the metaphase plate while the other bivalents are still on it. One such cell is photographed in *Fig.* 347. These chromosomes are taken to be the precociously separating X and Y, and it is concluded that the testis tissue has the XY constitution normally found in the male.

FIG. 339.—Ovary, showing primordial follicle below and part of late ripening follicle above. H. and E. (× 90.)

It was not possible to make a chromosome count in the ovarian tissue. Many dividing cells were obtained by scraping the inside of follicles, but these cells are very small.

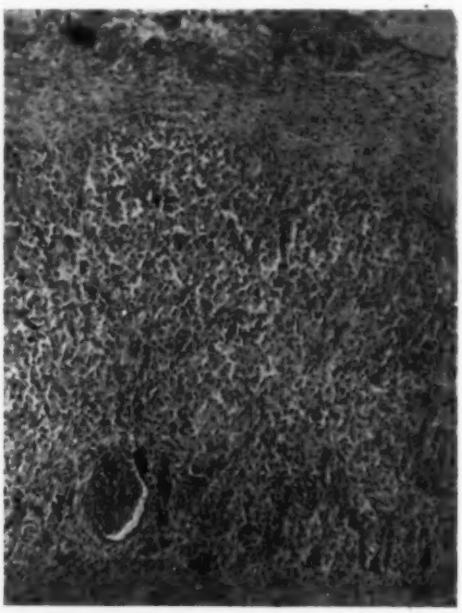

FIG. 341.—Corpus luteum showing organized inner border above, granulosa-lutein layer in centre, small darker theca-lutein leashes and dilated venules below. H. and E. (× 60.)

Sections of the testis showed in some tubules a number of cells which have heavily Feulgen-staining irregular nuclei which appear pyknotic. These cells occupy positions in which second meiotic divisions and spermatids would be expected.

DISCUSSION

Pathological and Cytological Findings.—The anatomical findings and the absence of hæmaturia suggest that the uterus consisted of an isolated corpus ending blindly below.

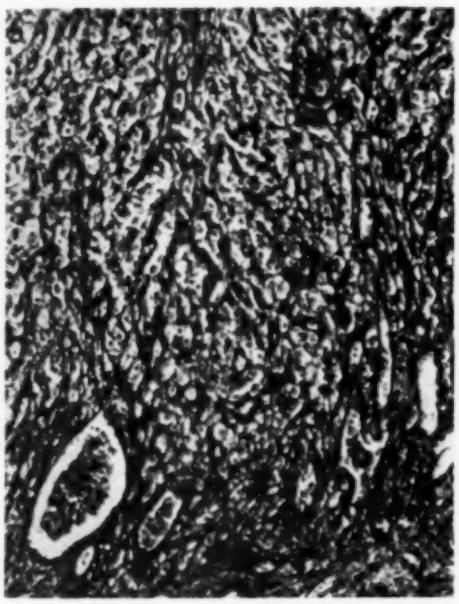

FIG. 342.—Reticulum impregnation of outer two-thirds of similar section to last at somewhat higher magnification. Modified Wilder. (× 105.)

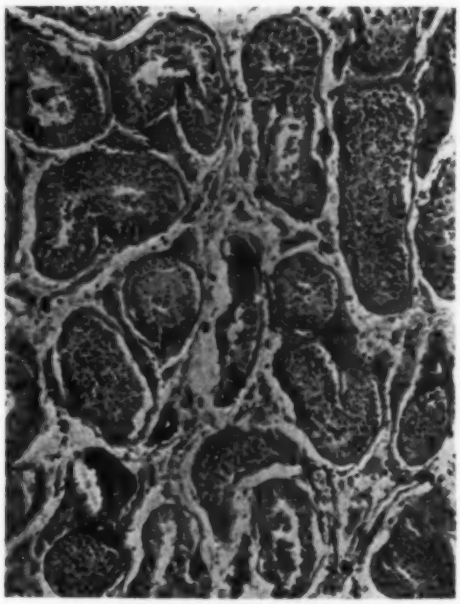

FIG. 343.—Testis, showing normal general appearance. Heidenhain's iron hæmatoxylin. (× 60.)

Both tube and ovary were essentially normal. The structures shown in *Fig.* 337 are the medullary canals or cysts, variously derived, in theory, by v. Franqué (1898) from mesonephric rests, by Meyer (1913) from remains of medullary cords, and by

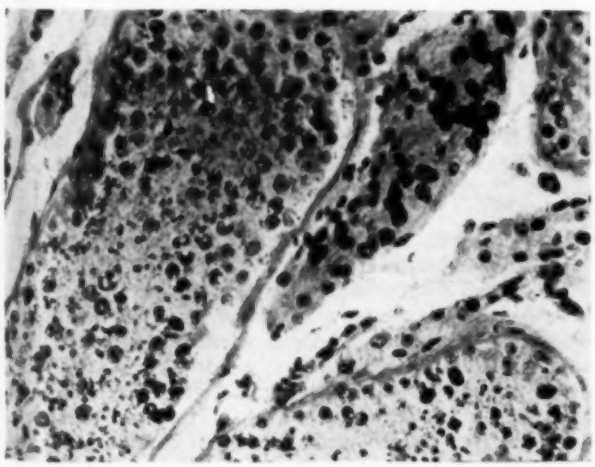

FIG. 344.—Right upper corner of last, showing numerous dividing spermatocytes and a group of Leydig cells with two mitotic figures. (× 187·5.)

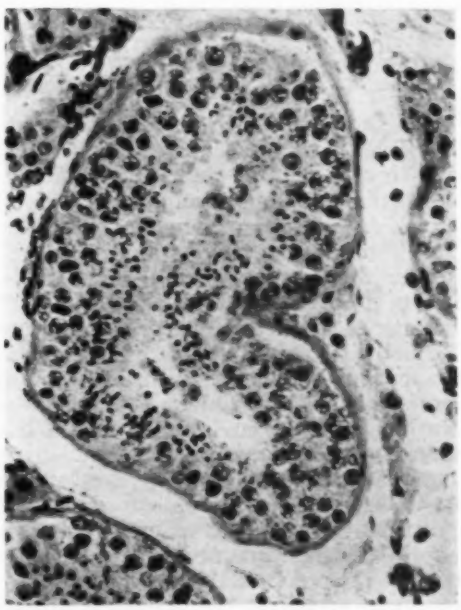

FIG. 345.—Tubule from left upper region of *Fig.* 341 showing nearly all spermatocytes in cell division and inner zone of (mainly) spermatozoa. A few radially elongated Sertoli-cell nuclei can be seen. (× 187·5.)

Joachimovitz (1935) from either rete or ovarian surface. A fifth possibility, put forward here, is an origin from follicles at the stage of a single epithelial granulosal ring, following degeneration of the ovum. This is suggested by the presence of such follicles in line with the 'tubules' in this case and the existence of forms transitional between the two seen by one of us (P. E. H.) in another ovary. The histogenesis may be variable: but even if, as Meyer suggested, these structures are homologous with the tubuli recti testis, they do not imply any special rudimentary bisexuality, for they are common in ovaries from normal women. It is of interest that the cells shown in *Fig.* 336 were present only in very modest numbers, for they are thought to be

homologous with the Leydig cells of the testis. The subject has recently been reviewed by Sternberg (1949).

The healthy majority of the testis tubules were compared with those of a normal testis from a man of 24 dying after a truck accident and on whom an autopsy was carried out a few hours later. No essential difference was seen save for a larger number of spermatozoa in the present case. The pyknotic elements present in some tubules suggest

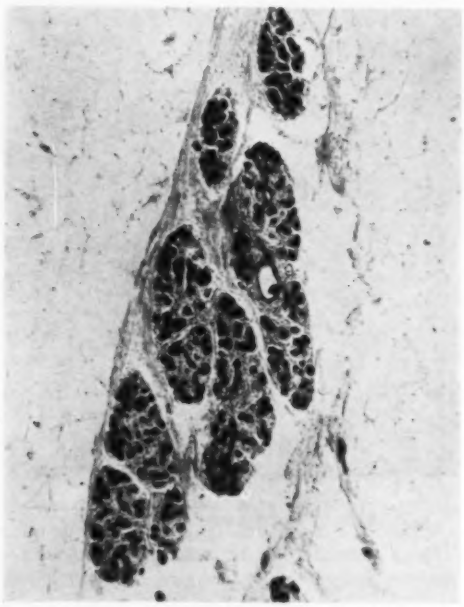

FIG. 346.—Part of breast lobule showing normal architecture. H. and E. (× 30.)

that some of the products of the first meiotic division are non-viable. One possible cause of such non-viability is a chromosome deficiency or duplication which is lethal in the haploid condition. The present observations do not give evidence of a chromosome structural abnormality which could be held responsible for the degenerate cells observed, but the possibility of a chromosome change is not excluded. The presence of the degenerate cells may thus indicate a genetic situation which might explain the sexually abnormal phenotype of this individual. However, it is equally possible that it reflects merely the abnormal hormonal environment of the tissue.

General.—The literature is reviewed by Young (1937) and Gudernatsch (1944): the latter found 37 cases of true hermaphroditism reported up to the end of 1944. Young used strict criteria, requiring microscopical demonstration of both ovary and testis. In particular he excluded cases (*a*) where testis had been microscopically demonstrated and ovary deduced from the presence of vaginal bleeding, and (*b*) where ovary had been microscopically demonstrated and testis deduced from the presence of spermatozoa in the seminal fluid. The first reservation appears to be justifiable, but not the second, nor has it been applied by later writers. The case of Urechia and Teposu (1933), excluded by Young, should therefore be restored to the list, and that of Malgras and Gricouroff (1939), included by Gudernatsch, should be discarded. This leaves the total number unaffected.

In addition, 4 acceptable cases published between 1937 and 1944 have apparently been overlooked, e.g. Ombrédanne (1939), Lindvall and Wahlgren (1940, Case 1), Brachetto-Brian, Grimaldi, and Costa (1943), and Jahier and Kehl (1943). Since 1944 7 acceptable cases have been published, viz., Davis and Scheffey (1946), de Moura and Basto (1946), Weed, Segaloff, Wiener, and Douglas (1947),

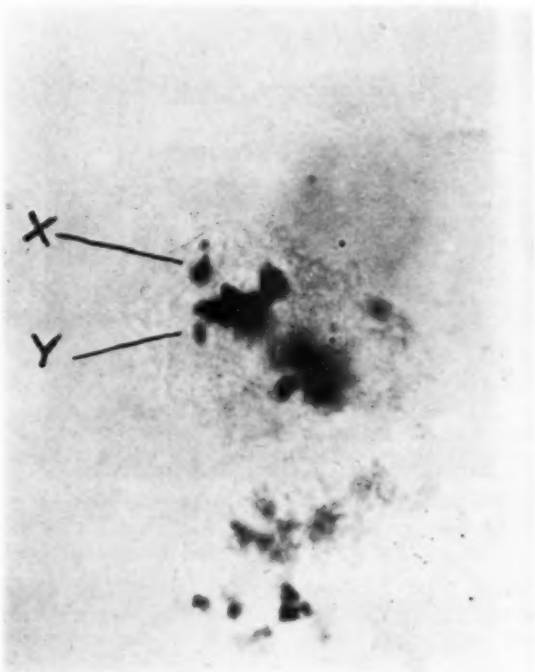

FIG. 347.—Feulgen squash of testis. First meiotic metaphase. In the upper cell two unequal elements are lying off the metaphase plate, the larger (X) near the upper pole, the smaller (Y) in the same part of the spindle, near the lower pole. (× 1500.)

Patton (1948), Stromme (1948), Rabadan, Ortiz, de la Peña, and Llusia (1950), and Capon (1951), making, with the present case, 49 in all. The list may be incomplete, however, as several papers possibly containing case reports are unavailable in London.

Since 1944 4 further supposed cases have been published which appear to be unacceptable, e.g., Portela and Fernandez-Ruiz (1945) and Haberkorn (1948)—no microscopy of testis or seminal fluid; Stirling (1946)—microscopy of testis unconvincing; and Shields, Freudenberger, and Ogilvie (1949)—ovary unconvincing. This doubt as to the diagnosis is more conventional than real. The literature contains many inadequately verified cases, some of which were probably genuine. The use of strict criteria, however desirable on other grounds, probably gives a misleading impression of the rarity of the condition and the prospect of encountering a case.

The 49 proven cases are made up as follows: lateral hermaphroditism (testis + ovary)—15 cases; unilateral hermaphroditism—16 cases (testis + ovotestis 5, ovary + ovotestis 11); bilateral hermaphroditism (ovotestis + ovotestis)—12 cases;

indeterminate (ovotestis + other gonad not sectioned)—6 cases.

Tables I and *II* show respectively the gonadal and other findings in the 15 published cases of lateral hermaphroditism. *Case* 12 differs from the rest in that the pelvic organs were mainly female, with a rudimentary uterus, tubes, and a pelvic testis in the site proper to the left ovary. The other 14 have more or less completely formed male and female internal organs on opposite sides. In Young's 2 cases uterus, tube, and ovary were found in a left inguinal hernial sac; in the remainder they were in the pelvis. Both external genitalia and bodily configuration have varied from almost normal male to almost normal female. Six cases have had external menstrual bleeding. Two cases (*Cases* 3 and 14) had regular attacks of abdominal pain, probably due to bleeding into a closed uterus, and our own case had menstruated in this way at least once. In only 2 post-pubertal cases (*Cases* 2 and 8) was there no apparent menstruation in association with an ovary containing ripening follicles.

Table I.—Age and Gonadal Findings in 15 Cases of Lateral Hermaphroditism

Case No.	Name and Date	Age	Testis				Ovary	
			Site	Size	Tubules	Interstitial Cells	Size	Follicles, etc.
1	Photakis, 1916	36	R. inguinal hernial sac	4 × 2 × 2 cm.	Involuted—no spermatozoa	Present	2 × 5 × 0·4 cm.	Primordial only
2	Young, 1924, 1937	18	R. scrotal sac	5 × 2·5 × 3 cm.	No spermatogenesis	Hyperplasia	5 × 3 × 2 cm.	All stages + fresh corpus luteum
3	Urechia and Teposu, 1933	25	L. scrotal sac	Normal	No biopsy. Living sperms in seminal fluid		Slightly reduced in volume	Follicles + corpora lutea and albicantia
4	Lindvall and Wahlgren, 1936	19	R. scrotal sac	3·5–4 × 2 × 1 cm.	Atrophic. No spermatogenesis	Scanty	Normal	All stages + corpora lutea
5	Raynaud, Marill, and Xicluna, 1939	18	L. scrotal sac	The size of a haricot	Involuted, but all stages of spermatogenesis claimed	Rare	5 × 2·5 × 2 cm.	Ripening follicles + corpus luteum
6	Young, 1940	20	R. scrotal sac	5·3 × 3 × 2·8 cm.	All stages of spermatogenesis	Rather scanty	1·6 × 1·5 cm.	None (stroma + psammoma bodies only)
7	Lindvall and Wahlgren, 1940, Case 1	18	R. inguinal hernial sac	Hazel-nut	Biopsy at age of 2 normal for this age		4 × 2 × 2 cm.	Ripening follicles
8	Brachetto-Brian, Grimaldi, and Costa, 1943	24	L. scrotal sac	No data	Sertoli cells + spermatogonia	Some increase	No data	Ripening follicles
9	McIver, Seaburgh, and Mangels, 1944 Case 2	17	L. scrotal sac	Normal	No biopsy. Motile sperms in seminal fluid		Not given. Micropolycystic in figure	Corpus luteum hæmatoma
10	McKenna and Kieffer, 1944, Case 2	3	R. scrotal sac	Normal for age	Immature	No data	10 × 8 × 4 mm.	No data
11	de Moura and Basto, 1946	19	L. scrotal sac	Normal	No biopsy. Sperms in seminal fluid		No data	Follicle cysts (+endometriosis)
12	Davis and Scheffey, 1946	6½	Pelvis, in site appropriate to left ovary	No data	Immature	No data	1·5 × 12 × 1 mm.	Doubtful 'primitive ova'
13	Patton, 1948	c. 20	R. labium majus	No data	Hypoplastic—no spermatogenesis	Normal	No data	Ripening follicles
14	Rabadan, Ortiz, de la Peña, and Llusia, 1950	19	L. scrotal sac	No data	Puncture showed immature sperms		Slightly enlarged and polycystic	All stages to corpus luteum (+endometriosis)
15	Present case	14	R. scrotal sac	Normal	All stages of spermatogenesis	Slightly increased	3·5 × 3 × 1·5 cm.	All stages to corpus luteum

The gonads might be expected to show the pathological effects of (*a*) œstrogens on the testis, and (*b*) androgens on the ovary. According to Burrows (1949) these are respectively (*a*) inhibition of spermatogenesis and interstitial-cell hyperplasia, and (*b*) enhancement of follicle maturation—with increase of ovarian size—by small doses, its suppression by large. All these effects (including micropolycystic ovaries) are represented in different cases in *Table I*. There is no clear correlation between age and failure of spermatogenesis. Our own case is the first in which substantially normal function of both gonads has been established by biopsy.

Little is known of the causes of human intersexuality. Aberrations in sexual differentiation might in theory depend upon: (1) An abnormal combination of sex-determining genes already present in the zygote; (2) An abnormal separation of sex chromosomes in mitosis at any time from the first cleavage till the onset of genital differentiation, resulting in a mosaic of male and female tissue; (3) Thereafter, until the completion of genital development, an

abnormal hormonal environment deriving from (*a*) the mother, (*b*) a twin, or (*c*) the embryo itself. It is reasonably certain that in 3(*c*) fall the cases of female pseudohermaphroditism associated with other findings in lower animals. The familial incidence noted with some intersexes does not necessarily place them in group 1: it is found also in cases with adrenal hyperplasia.

Table II.—Other Findings in 15 Cases of Lateral Hermaphroditism

Case No.	Name and Date	External Menstrual Bleeding	Social Sex	Breasts	Bodily Habitus	Genital Tubercle	Labioscrotal Folds	Urethra	Müllerian Duct	Wolffian Duct
1	Photakis, 1916	—	F.	M.	Male	Penis	Scrotum	Penoscrotal hypospadias	L. tube, bicornuate corpus, cervix, urethral vagina	Normal epididymis and vas
2	Young, 1924, 1937	—	M.	M.	Male	Penis with chordee	Scrotum	Penoscrotal hypospadias	L. tube + uterus	Epididymis abnormal. No vas
3	Urechia and Teposu, 1933	—	M.	F.	Male, with female subcutaneous fat	Penis	Scrotum	Penoscrotal hypospadias	R. tube + uterine corpus	No data
4	Lindvall and Wahlgren, 1936	+	M.	F.	Mainly male	Penis	Scrotum	Scrotal hypospadias	L. tube, corpus, cervix, connecting with urethra	Apparently normal epididymis and vas
5	Raynaud, Marill, and Xicluna, 1939	+	M.	F.	Mixed, mainly female	Penoclitoris 2 cm.	L. scrotal sac; R. labium majus	Hypospadias	R. tube, uterus, urethral vagina	Epididymis palpable
6	Young, 1940	—	M.	M.	Male	Penis with chordee	Bifid scrotum	Scrotal hypospadias	L. tube, corpus, cervix, vagina, attached to prostate	Thin elongate epididymis. Normal vas
7	Lindvall and Wahlgren, 1940, Case 1	+	M.	F.	Mainly male	Penis, 4 cm.	Scrotum continued anteriorly like labia majora	Penoscrotal hypospadias	L. tube, bicornuate corpus, cervix, urethral vagina	No data
8	Brachetto-Brian, Grimaldi, and Costa, 1943	—	M.	R.-F. L.-M.	Mixed	Penis	Scrotum	Hypospadias	R. tube + isolated unicorn uterus	Epididymis and vas normal
9	McIver, Seaburgh, and Mangels, 1944, Case 2	+	M.	F.	Mixed, with broad pelvis	Penis	Scrotum	As normal male	R. pus-tube + uterus communicating with urethra	No data
10	McKenna and Kieffer, 1944, Case 2	N.A.	M.	N.A.	Mixed, mainly male	Penoclitoris	R. scrotal sac normal; L undeveloped	Hypospadias	L. tube, corpus, cervix, urethral vagina	No data
11	de Moura and Basto, 1946	+	M.	F.	Mainly male	Penis	Scrotum	As normal male	R. tube + normal corpus and cervix	Epididymis felt normal
12	Davis and Scheffey, 1946	N.A.	F.	N.A.	Muscular	Penis	Labia majora	Perineal hypospadias	Tubes, rudimentary uterus, urethral vagina	No data
13	Patton, 1948	+	M.	F.	Female	Penoclitoris	R. labium majus; L. labioscrotum	Hypospadias	L. tube, corpus, cervix, vagina opening into vestibule	No data
14	Rabadan, Ortiz, de la Peña, and Llusia, 1950	—	M.	F.	Female	Penis, 4 cm.	Scrotum with anterior hood on L.; nil on R.	Penoscrotal hypospadias	R. tube, unicorn corpus with cervix	Epididymis and vas normal
15	Present case	—	M.	F.	Female	Penis	Scrotum	Hypospadias	L. tube + isolated unicorn corpus	Epididymis and vas normal

adrenal hyperplasia, that 3(*b*) is void in man, and that 3(*a*) contains a single case, described by Brentnall (1945), in which a woman with a masculinizing tumour of the ovary during pregnancy gave birth to an intersexual fœtus, probably a female pseudohermaphrodite. The disposition of the rest is a matter of guesswork prompted by experimental and

Greene (1944) advanced the hypothesis that genetic processes initiate the chain of reactions which cause differentiation between male and female structures. The genes determine whether the gonad shall be an ovary or a testis. The ovary or testis then produces hormones which cause other hitherto undifferentiated organs to develop towards masculinity

or femininity. He doubted whether any single hypothesis could account for all aberrations from this plan and suggested that the fault in persons with a separate ovary and testis is essentially genetic; and that in those with an ovotestis the cause is hormonal. Also hormonal, in his view, are the so-called pseudohermaphrodites, in which the genetic pattern of one sex has been altered by the interference of abnormal hormonal influences (as, for example, in the freemartin and in intersexuality due to adrenal tumours). Moore (1947), on the other hand, believed that the development of the ducts is genetically determined *ab initio*. Jost (1947) showed that when very young rabbit embryos are castrated before the initiation of sexual differentiation they all develop along female lines. Similar observations are recorded for mice (Raynaud and Frilley, 1947). This might be due either to an initially feminine bias or to the action of maternal œstrogens unopposed by embryonic androgens. The experiments therefore do not settle the question. In any case they apply only to rodents and take no account of the great differences in developmental and hormonal patterns between rodents and human beings.

A striking feature of all but one of the 15 cases of lateral hermaphroditism is the perfect asymmetry of development not only of the gonads but also (so far as data are available: *see Tables I, II*) of their associated ducts. Further, Brachetto-Brian and others (1943) describe a patient with a right shoulder higher, and right arm shorter, than the left, right-sided gynæcomastia, right hip and buttock fuller than left, right foot smaller than left, and conclude that right and left sides of the body are here respectively masculine and feminine. There are no other reported cases with a similar general asymmetry, but 4 cases have shown asymmetric development of the labioscrotal folds (*see Table II*).

As it is hardly easier to explain this in terms of genes present in the zygote and distributed to every cell in the body than in terms of hormones, several writers have suggested an abnormal distribution of sex chromosomes early in embryonic life. There are three possibilities: (1) The zygote is XY and non-separation of two daughter X chromosomes in a division subsequent to the first gives rise to an XXY cell (and a complementary non-viable Y cell). Part of the body may then have an XXY (female) constitution and part an XY (male). (2) The zygote is XXY and one X chromosome is lost from part of the body. (3) The zygote is XX (as in a normal female), and the loss of one X chromosome results in part of the body being XO. This assumes that XO tissue is viable. There is still some difference of opinion as to whether the human male is normally XY or XO. Matthey (1949) in summing up the evidence states that, while in his view the Y chromosome is present, the matter cannot be looked upon as finally settled.

In the present case the testis appears to be XY, and we must assume that the zygote possessed a Y chromosome. It is unfortunate that we were unable to determine the chromosome constitution of the ovarian tissue, and that we thus cannot offer any evidence for or against the notion that lateral hermaphroditism is due to a mosaic condition of the sex chromosome constitution.

We know of 2 previous studies of the chromosomes in human intersexes, viz., Severinghaus (1942) and Witschi and Mengert (1942). Severinghaus gives a detailed account and figures of the XY complex found in a male pseudohermaphrodite of male general appearance, with a hypospadic penoclitoris and labial testes. In a rather similar case Witschi and Mengert failed to confirm this, but in the absence of details or illustrations their findings carry little conviction. It seems at least clear that not all subjects with pronounced genital malformations are modified females.

Further studies on human intersexes of all kinds seem called for, and it is worth noting that these are quite feasible and can only be made on promptly and appropriately fixed human surgical material. Small pieces of tissue should be taken, placed for 24 hours in a mixture of absolute alcohol 3 parts and glacial acetic acid 1 part, and then transferred to 70 per cent alcohol. (Many other fixatives can be used: this one has the merit of simplicity.) Testis, ovary, and endometrium should be taken where available, but as these may prove unsuitable it is advisable also to remove a lymph-node: this should be cut across before fixation.

SUMMARY

A case of true lateral hermaphroditism is described.

The pathological and histological characters of the ovary, tube, uterus, testis, and breasts are given, including the chromosome constitution of the testis.

Hermaphroditism is discussed and the literature reviewed. The main features of 14 published cases of true lateral hermaphroditism are set out for comparison with those of the present case.

The therapeutic problems and surgical management of the present case are described.

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CALCULI IN THE APPENDIX

BY L. P. CLARK
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A TRUE calculus, as opposed to a fæcolith, in the appendix is not an uncommon condition, but its incidence appears to vary widely in the experience of various writers. Even when radiological evidence is strongly suggestive, the diagnosis is frequently overlooked, and for this reason it is thought that report of a further case may be useful.

Moloney (1947) reports a case of a gangrenous appendix containing a large calculus which, on its radiological appearance, was considered to be a gall-stone in the ileum.

Felson and Bernhard (1947) reviewed 100 cases reported in the literature from 1900 and presented 10 further cases occurring in 300 appendicectomies over a period of three years. Since then, 19 more cases have been recorded in various reports. Since 1900 there have been 8 references to calculi in the appendix in British journals.

In the total of 129 cases mentioned above, all but 13 presented clinical or surgical findings of acute appendicitis. The following description is of a case of calculi occurring in an appendix not acutely inflamed.

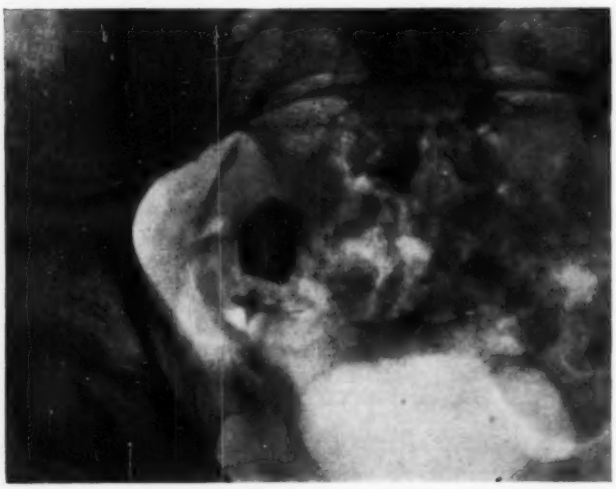

FIG. 348.—Intravenous pyelogram. Calculi distinct from lower end of right ureter.

FIG. 349.—Appendix opened to show the larger calculus. (× ½.)

CASE REPORT

A man, aged 35, attended hospital on Nov. 13, 1950, with a history of attacks of abdominal pain and constipation for five years, each attack lasting a week and occurring twice in the previous three months. The pain was generalized in the abdomen, continuous in type, not severe, and not accompanied by nausea or vomiting. Otherwise, he was in good health and had had no previous illnesses of any significance. Examination showed him to be of normal development and healthy in appearance. The abdomen was not tender and the only viscus palpable was the descending colon. The other systems revealed no abnormality on examination.

A very tentative diagnosis of chronic appendicitis was made and investigations were instituted. A barium-meal examination revealed no abnormality of the alimentary tract, except that the appendix did not fill. Two opacities were shown in the region of the right sacro-iliac

joint. An intravenous pyelogram proved that the opacities were clear of the right ureter (*Fig.* 348). The larger opacity showed a peripheral increased density suggestive of lamination and not the mottling characteristic of calcified glands.

The patient was admitted to hospital and laparotomy performed on Jan. 23, 1951. The appendix was found to be free, lying across the pelvic brim, and thickened and firm to palpation. The proximal half was distended to a bulbous shape by the contained calculi. There were no other visible changes indicative of past or present inflammation. The appendix was removed by the purse-string technique. Exploration of the abdomen revealed

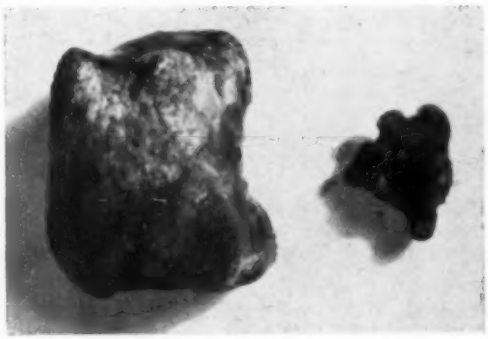

FIG. 350.—Calculi from the appendix. (× 2.)

no other abnormality. Following operation the patient made an uninterrupted recovery.

Examination of the appendix (*Fig.* 349) showed its wall to be markedly thickened. There were two hard calculi in the lumen, the proximal one cuboidal in shape, light brown in colour, and a smaller star-shaped calculus, almost black in colour (*Fig.* 350). The wall of the appendix at the site of the larger calculus was thinned out, but there was no ulceration of the mucosa.

PATHOLOGY.—The report on the appendix and calculi is as follows :—

Appendix : Section of the proximal end showed an intact mucosa and a well-developed muscularis, with no histological evidence of past or present inflammatory changes in the submucosa or subserosa.

Calculi : The fragments submitted for analysis weighed 0·25 g., were pigmented a light brown colour, due to stercobilin, and were moderately soft in texture. The material consisted mainly of calcium phosphate. There was also present 10 per cent total fat but only a trace of cholesterol—less than 1 per cent.

COMMENTARY

Aetiology.—It is generally considered that inspissation of fæces in the appendix produces a fæcolith which irritates the mucosa to cause a low-grade catarrhal inflammation with secretion of mucus. Inorganic salts in the mucus are precipitated on the surface of the fæcolith. Recurring attacks of such a process will result in a laminated stone. Foreign bodies may form the nidus for a stone and rare cases have been reported of gall-stones finding their way into the appendix.

Incidence.—This appears to vary widely. Bunch and Adcock (1939) found one case of stone in over 2000 patients with appendicitis, while Steinert, Hareide, and Christiansen (1943) recorded 10 cases in 104 appendicectomies. The case reported here is the first met with in 500 appendicectomies. No doubt many fæcoliths contain calcium phosphate, insufficient to cast a shadow in the plain radiograph through the abdominal wall of an adult. The thinner abdominal wall in children may not mask them so completely, and Childe (1947) records 8 cases occurring in children over a period of 11 years.

Diagnosis.—Felson and Bernhard (1947), in their review of 110 cases, found that there were no characteristic clinical findings, the diagnosis depending entirely on X-ray appearances. They state that a laminated calculus showing in the right lower quadrant of the abdomen will almost invariably prove to be an appendicular stone. Absence of lamination favours a ureteric calculus which can be differentiated by a pyelogram, while calcified lymph-glands show a fragmented appearance. A cholecystogram should eliminate the possibility of an inferiorly displaced gall-bladder containing stones, as over 60 per cent of recorded cases of appendicular calculi have occurred in males below the age of 40. Other conditions which have to be borne in mind are phlebolith, calcified appendix epiploica, and barium residue.

Treatment.—In the 129 cases of appendicular calculi reported since 1900, 116 presented with an attack of acute appendicitis, in 50 per cent of which perforation had occurred. In 2 of these cases a symptomless calculus had previously been demonstrated by X rays as an incidental finding. Both cases subsequently developed acute appendicitis.

It would, therefore, appear reasonable to conclude that radiological demonstration of a stone in the appendix is a strong indication for appendicectomy, irrespective of the presence or absence of symptoms. Should mild symptoms of appendicitis be present, it would be wise to advise appendicectomy as soon as possible rather than admit from a waiting list.

In a case of acute appendicitis in which the diagnosis is in doubt, a plain radiograph of the abdomen would prove of value should an appendicular calculus be present.

SUMMARY

A case of calculi in the appendix is described.

The diagnosis of the condition rests upon radiological appearances, which are fairly characteristic.

The condition is an indication for early appendicectomy.

I wish to thank Dr. Knowles, of the Southern Group Laboratory, for the pathological reports, and the Department of Medical Photography, Guy's Hospital, for the photographs.

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LATE RESULTS OF COMPOUND FRACTURES OF THE TIBIA IN BATTLE CASUALTIES TREATED IN A PENICILLIN UNIT

By M. E. FLOREY, J. S. JEFFREY, R. W. N. L. ROSS, and E. C. TURTON

This paper describes briefly the processes of repair undergone, and the results obtained up till 6 years from wounding, in 40 consecutive cases of compound comminuted fractures of the tibia received in a Unit* set up by the Medical Research Council Penicillin Trials Committee, at the Radcliffe Infirmary, Oxford, in August, 1944.

As the accommodation available for the Unit was limited, the trial was confined to seriously injured casualties. Accordingly arrangements were made with a nearby receiving aerodrome to select such cases and to forward them to the Unit. It was thus hoped to provide a severe enough test of the power of penicillin so to control major sepsis that it was justifiable to take exceptional steps to secure early union and, in some cases, forego amputation, provided the limb had a sufficient blood-supply. It was planned to keep each man until his wound was healed and he was able to walk unaided—except for appliances needed for neurological lesions—or until events proved that his limb was of no use to him.

All the men were admitted between Aug. 1 and Dec. 1, 1944, 35 of them between the first and fifth day after wounding, the remaining 5 from the eighth to the nineteenth day. Few had a single wound—various soft-tissue wounds, nerve and vascular injuries, fractures elsewhere, and head injuries being among the other lesions which had to be attended to. Nearly all had had penicillin from the first day, the early stages of shock had already been dealt with, and débridement and splinting in plaster-of-Paris had been carried out as well. In consequence the problem for the Unit was to supervise the stages of repair through which each man had to pass before a decision might be made as to whether a useful limb could be restored to him.

On the arrival of the patient the wounds were inspected and specimens collected for bacteriological examination before penicillin was recommenced and the definitive operation performed. A minimal amount of apparently necrotic tissue and skin were removed and loose pieces of bone and foreign bodies extracted; after re-alinement, padded plaster was applied and windows cut over the wounds. An attempt was made to provide skin covering for all exposed bone at this time by means of undercutting and rotation flaps but the tension was often great. Thirteen wounds were closed and no further operation was necessary, but the remainder persisted as open wounds and required many sequestrectomies, much plastic repair and bone grafts, so that as many as 16 operations were required in the most severely injured patient.

CHEMOTHERAPY

Considerations of space prevent a detailed description of the methods of chemotherapy. Briefly, penicillin was the only systemic agent employed during the first 10 months of the Unit's existence and the dosage was low—15,000 units 3 hourly by intramuscular injection, or 100,000 units daily by intramuscular drip. It was used originally until sensitive organisms had disappeared from the wounds and later as a cover for every operation performed.

On the other hand, various drugs were freely applied locally in an effort to dispose of the Gram-negative bacteria which infested the wounds.

No operation was performed without careful preparation of the field in which the surgeon was going to work. Bacteriological examinations were made regularly once or twice a week throughout in order to determine, as far as possible, the appropriate application to be given daily till operation so as to control the prevailing bacteria.

At each operation not less than 100,000 units of undiluted penicillin powder or crystalline material were dispersed in the wound. Those patients requiring a number of operations also received streptomycin after 1945.

These precautions were taken to prevent possible infection by Gram-positive bacteria after operation, and unsatisfactory results caused by the ever-present Gram-negative bacteria.

BACTERIOLOGICAL FINDINGS

From the bacteriological studies the following information was found:—

Clostridia, including *Cl. Welchii*, were isolated from 26 wounds on admission, but these disappeared under treatment within the first 6 weeks; thereafter they were found only occasionally up to the end of 4 months.

Staph. aureus was originally found in 10 wounds, but disappeared in all but one instance, where a relatively resistant organism persisted and caused much damage to a bone graft until the screws were removed and local instillations of penicillin solution, bringing high concentrations to the site of the fracture and continued for many months, controlled the infection and a firmly consolidated bone was obtained. (For illustrations *see* Florey, 1952.) *Staph.*

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* The Unit originally consisted of Lt.-Col. James Jeffrey, as surgeon; Dr. Audrey Smith and later Dr. R. W. N. L. Ross as bacteriologists; Dr. C. Turton, registrar; and M. E. Florey as research assistant. The Sir William Dunn School of Pathology supplied accommodation and advice for the bacteriological work.

At the end of 6 months the Unit came under the supervision of the Radcliffe Infirmary Accident Service in the care of Sir Max Page and his assistants, Mr. Lazlo and Mr. Dennis Dunn, while plastic surgery was performed by Professor Pomfret Kilner and the bacteriology was supervised by Dr. R. L. Vollum.

In 1946 the remaining patients were transferred to Stoke Mandeville Ministry of Pensions Hospital where the final stages of surgical repair were carried out by Professor Pomfret Kilner and Mr. J. P. Armstrong, and where the Public Health Service Laboratory performed the bacteriology.

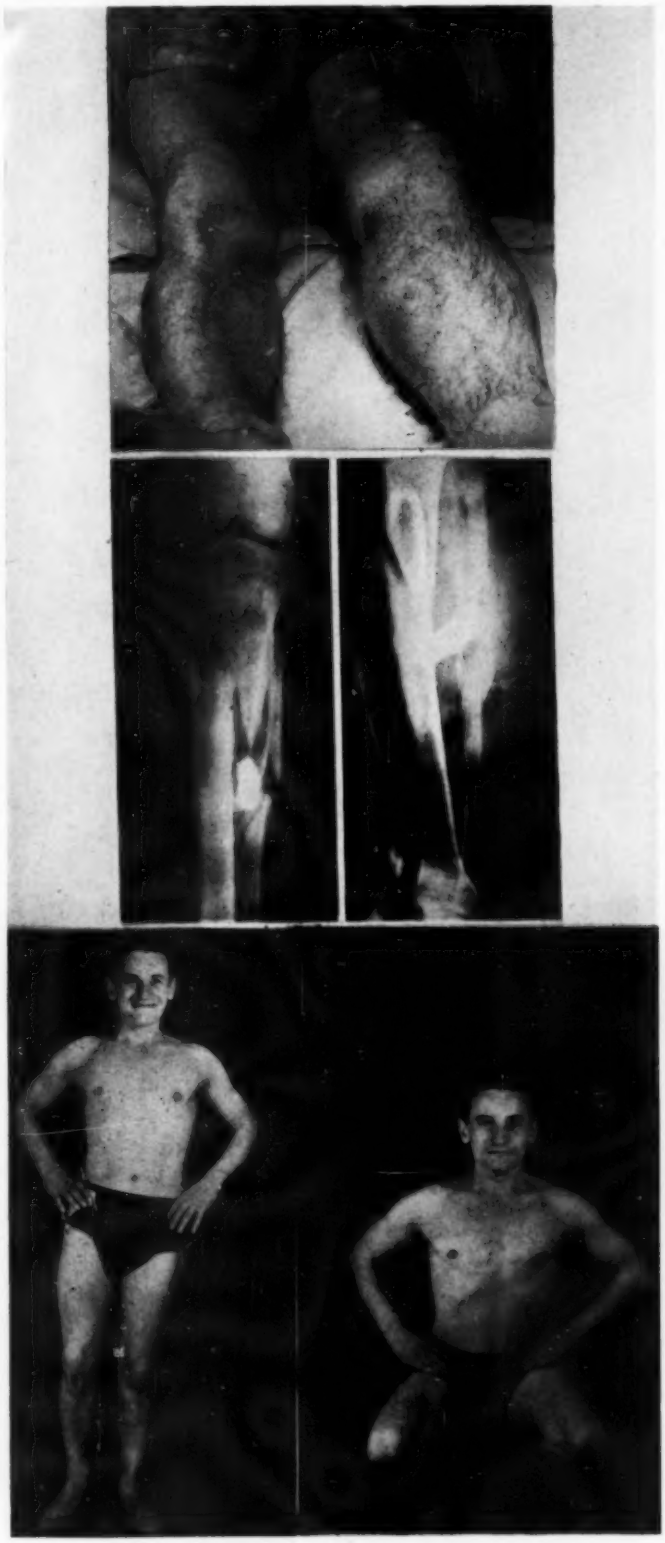

FIG. 351.—A, Fracture of left tibia—photograph 6 days from wounding. The left leg is very much more swollen than the right. The external wound was small but the damage to muscle was considerable. *Cl. Welchii* was isolated from the wound, and gas escaped on the removal of a foreign body from the site of the fracture; B, Radiographs of fracture taken 2 days before A, showing shattered tibia and fibula, with a foreign body lodged at its site. C, Functional result before discharge at 9 months: the patient can take his whole weight on the injured limb and flexion of knee and ankle are nearly normal.

FIG. 352.—A, Radiographs of fractured right tibia and fibula 5 days from wounding (no photograph of the wound was obtained); B, Radiographs 3 months later showing the many separating bony fragments at the site of the injury. Ten were removed at different times but a small sinus persisted intermittently—the patient, however, preferred to put up with the sinus rather than spend any more time in hospital; C, Functional result before discharge at 16 months. Free movement of joints above and below the site of fracture is seen.

FIGS. 351-353.—*Compound fractures of the tibia in battle casualties (upper third).*

FIG. 353.—A, Photograph of 2-day-old wound. The head of the tibia had been gouged out and no stability to the limb was given by the fibula—the limb is supported by hands below the back sheet; B, Radiographs taken on the same day; C, (*left*) Functional result 18 months later—the knee is ankylosed but there is good ankle movement and the patient can walk smartly and board buses without difficulty; (*right*) A close-up photograph of the plastic repair effected by an abdominal pedicle graft. The original deep wound has been well covered by skin and no sinus remains. (*From* Florey, 1952. By courtesy of the Oxford University Press.)

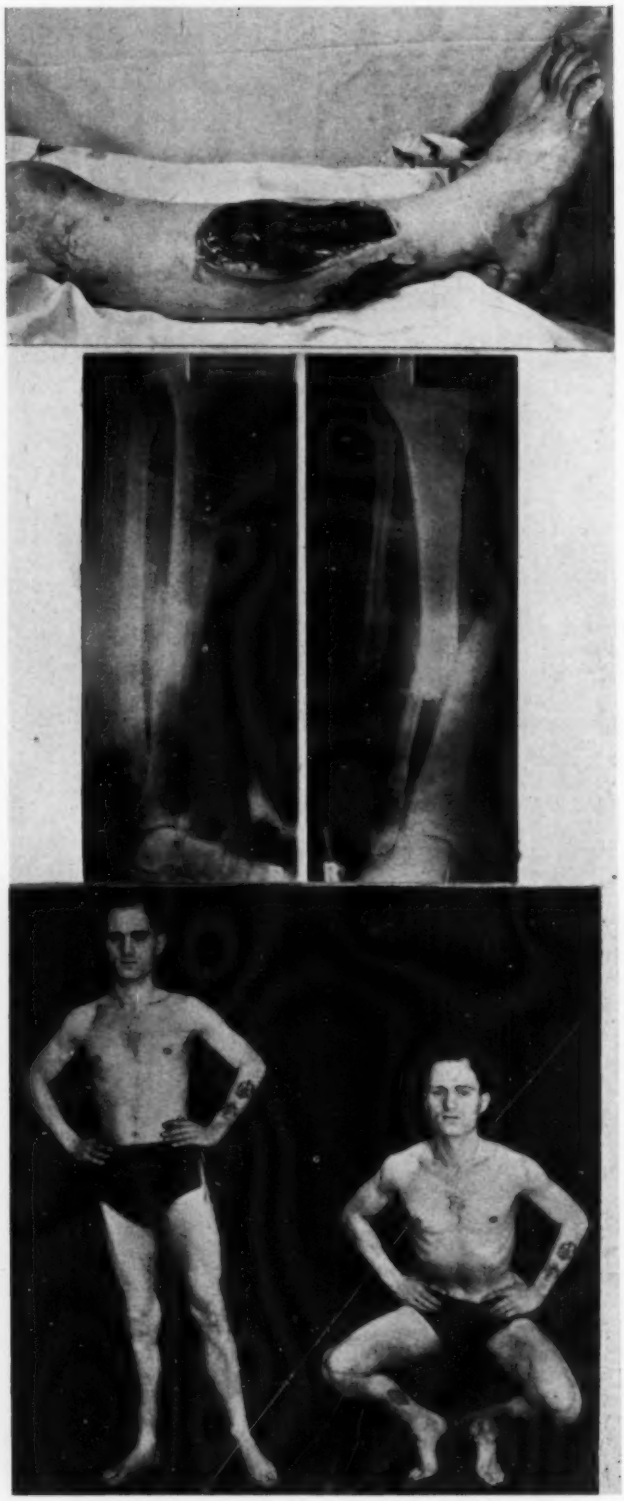

FIG. 354.—A, Photograph of 5-day-old wound with much muscle destruction; B, Radiographs of fracture at the same time; C, Functional result 15 months later when weight-bearing and flexion and dorsiflexion of knee and ankle are sufficient for usual purposes.

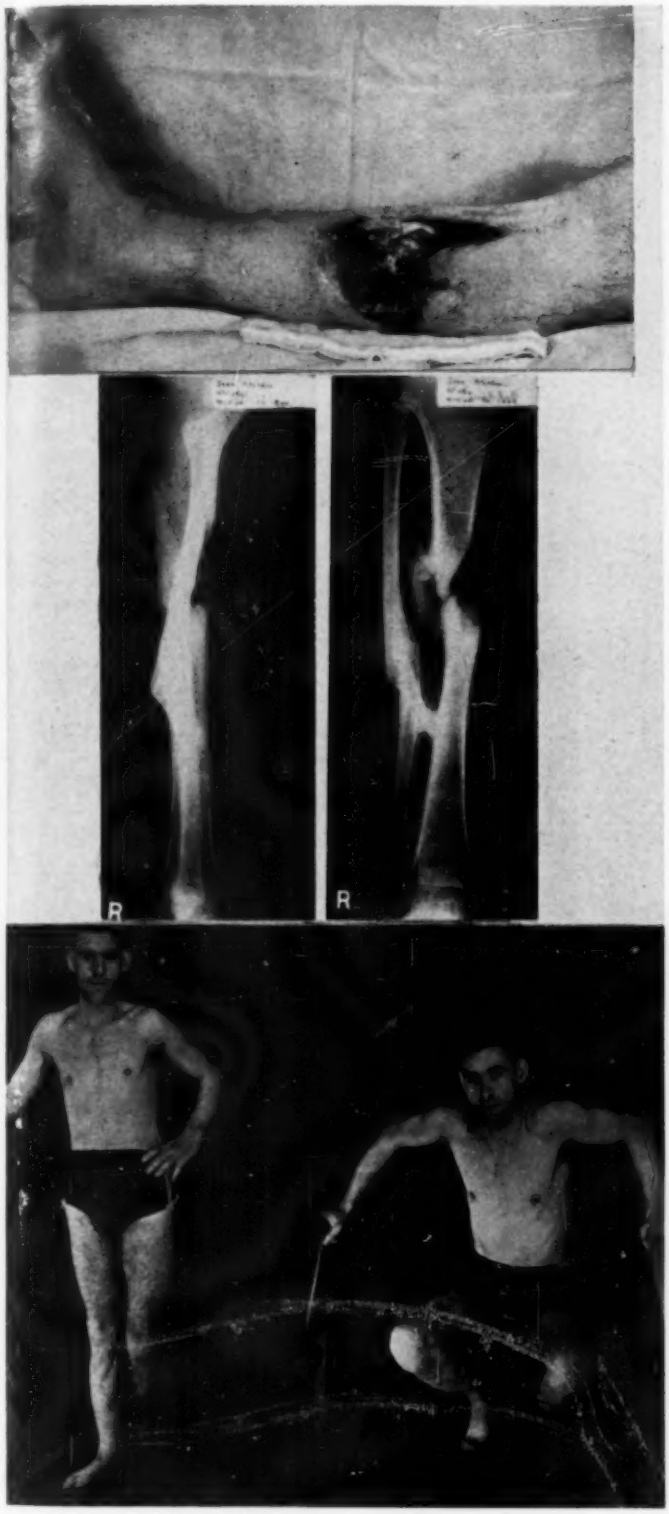

FIG. 355.—A, Photograph of wound 3 weeks from injury with bony fragments exposed. This man had had part of his other limb blown away and immediate amputation performed, making retention of this one imperative: B, Radiographs of fracture site 10 months after injury, after part of the exposed and dead tibia had had to be chiselled away and before a bone-graft was inserted: C, Functional result 16 months after injury—the skin covering was made by transfer of an abdominal flap before bone-grafting. There is no limitation of knee flexion or dorsiflexion of ankle.

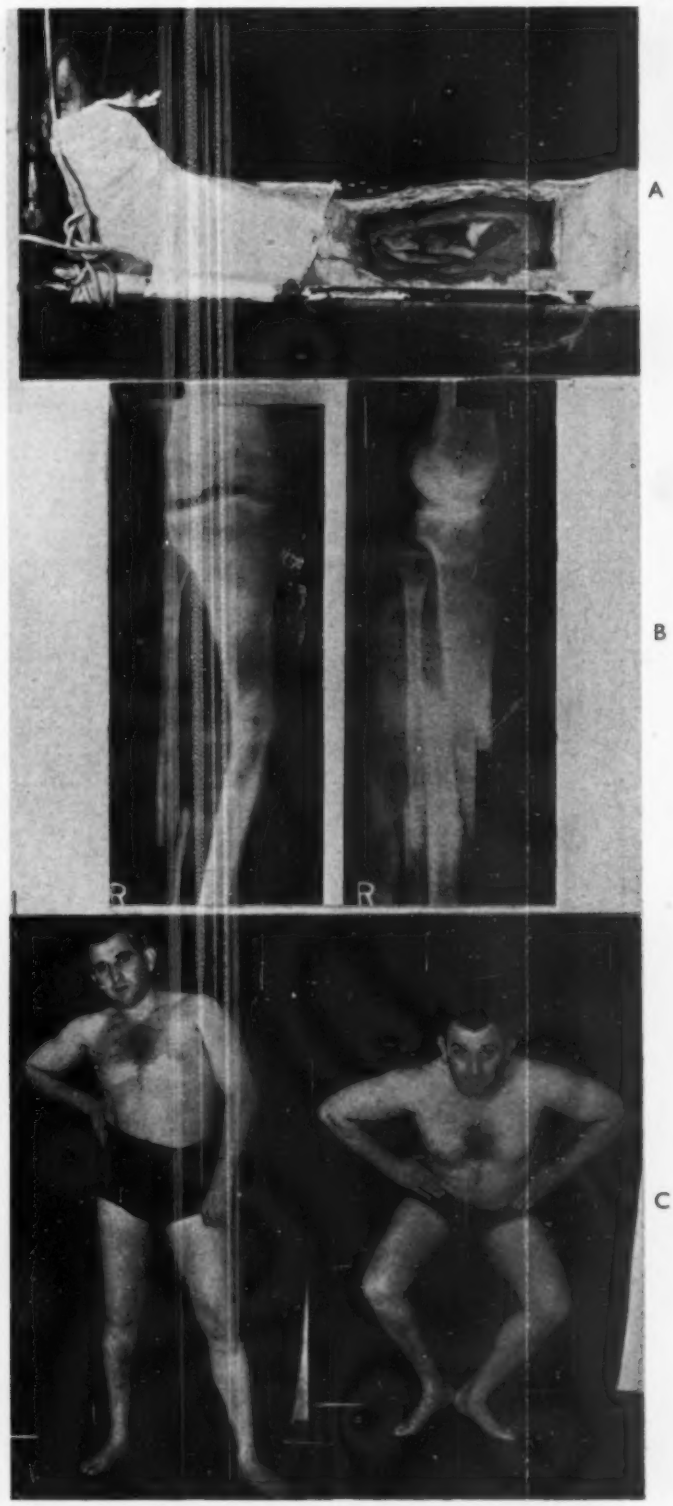

FIG. 356.—A, Photograph of wound 2½ weeks after injury. A separated fragment of tibia is exposed besides the upper and lower ends of the bone proper. This eventually united in part but a bone-graft was needed to replace the part that had died and been chiselled away: B, Radiographs taken at the same time; C, Functional result after 3 years. This man constantly complained of pain in his leg and radiographs showed repeated evidence of osteitis; it is possible that this damage to the bone was associated with the presence of a relatively resistant non-hæmolytic streptococcus which did not disappear till penicillin was applied locally—100,000 units daily. The poor functional result is in contrast with those of the other men, in whom osteitis was not suspected.

FIGS. 354-356.—*Compound fractures of the mid third of the tibia in battle casualties.*

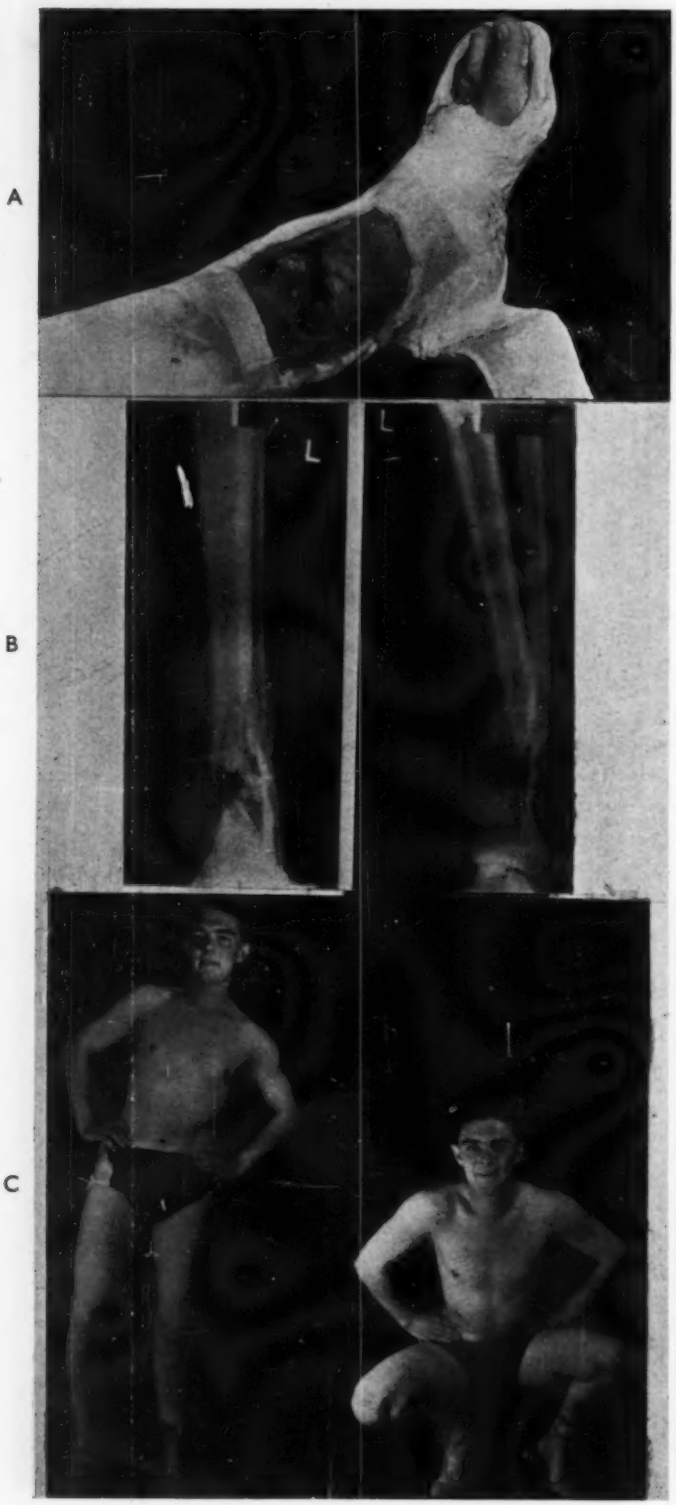

FIG. 357.—A, Wound 2 weeks from injury with bone exposed and little soft tissue to cover it; B, Radiographs taken at the same time; C, Functional result 10 months later—the bone has thickened considerably, not needing any graft to support its inner side. There has been no difficulty in weight-bearing but some limitation of ankle movement and intermittent dermatitis around the site of fracture where no pedicle graft was attached to it.

FIG. 358.—A, Wound 2 weeks from injury with exposed tendon. Pus is dripping into the receiving bowl; B, Radiographs of fracture at the same time with many small foreign bodies and much splintering of the bones; C, Functional result after 12 months. The ankle is fixed but the knee flexes fully and unaided weight-bearing is possible. A sinus discharges intermittently at the outer side of the leg, from which much dead bone has been scraped.

FIGS. 357-359.—*Compound fractures of the lower third of the tibia in battle casualties.*

aureus appeared frequently during healing but penicillin was applied immediately its appearance was reported and before any signs of inflammation were visible.

Penicillin-resistant staphylococci did not appear often until the wounds were reduced to sinuses, nor did they cause anything but superficial damage. When an epidemic arose, their origin was traced by phage typing to the hands of a dresser who had lately joined the Unit. Their number was reduced by undiluted penicillin locally applied each day, but they were not permanently eliminated.

Hæmolytic streptococci were seldom detected and, when tested, were not often found to be *Streptococcus pyogenes*.

Non-hæmolytic streptococci appeared intermittently but in 4 wounds were isolated from over half the swabs taken during the first three months after wounding. In 2 the radiologist reported signs of osteitis and the men continually complained of pain in their legs. Eventually removal of dead bone was required. Though the limbs ultimately united with the assistance of bone grafts, the movements of knees and ankles were limited for a much longer time than in other men. The third patient, in whom there was much destruction and sloughing of the anterior muscles of the leg for 2 months, was left with a small but frequently breaking down area of skin over the crest of the tibia but no obvious sequestrum. The fourth suffered from a crush injury involving his ankle joint and much dead bone was frequently scraped out from the bony cavity left there.

Dead tissue was repeatedly removed from all these patients, but whether non-hæmolytic streptococci caused or were merely a concomitant of dead tissue remains in doubt.

Gram-negative organisms, mainly coliforms, pseudomonas, and proteus, eventually appeared in all that had not healed by the sixth week in the Unit and in spite of the efforts made to observe the Medical Research Council's recommendations (1941) on a non-touch technique. Their clinical significance appeared to lie mainly in their ability to dissolve blood-clot formed after operation and so remove the bed in which granulations and subsequent fibrous and bony tissue might develop. This and their treatment were discussed in a separate paper (Florey, Ross, and Turton, 1947).

The accompanying illustrations (*Figs.* 351–360) are considered as representative of the types of wounds and fractures which were dealt with and the functional results before discharge from the Unit.

FOLLOW-UP

Six years after wounding it was possible to get in touch with 36 of the men; the remaining 4, being among those least severely injured, were discharged back to the Army for active service and unfortunately lost sight of. It is unlikely therefore that they lost their limbs on account of the injury for which they were treated and it can be reasonably claimed that 38 of the 40 had retained their limbs.

The two patients whose limbs were eventually amputated deserve more detailed study. The first had a fracture of the tibia at the level of the internal malleolus involving the ankle joint and a crushed astragalus. The fracture united but the foot was in

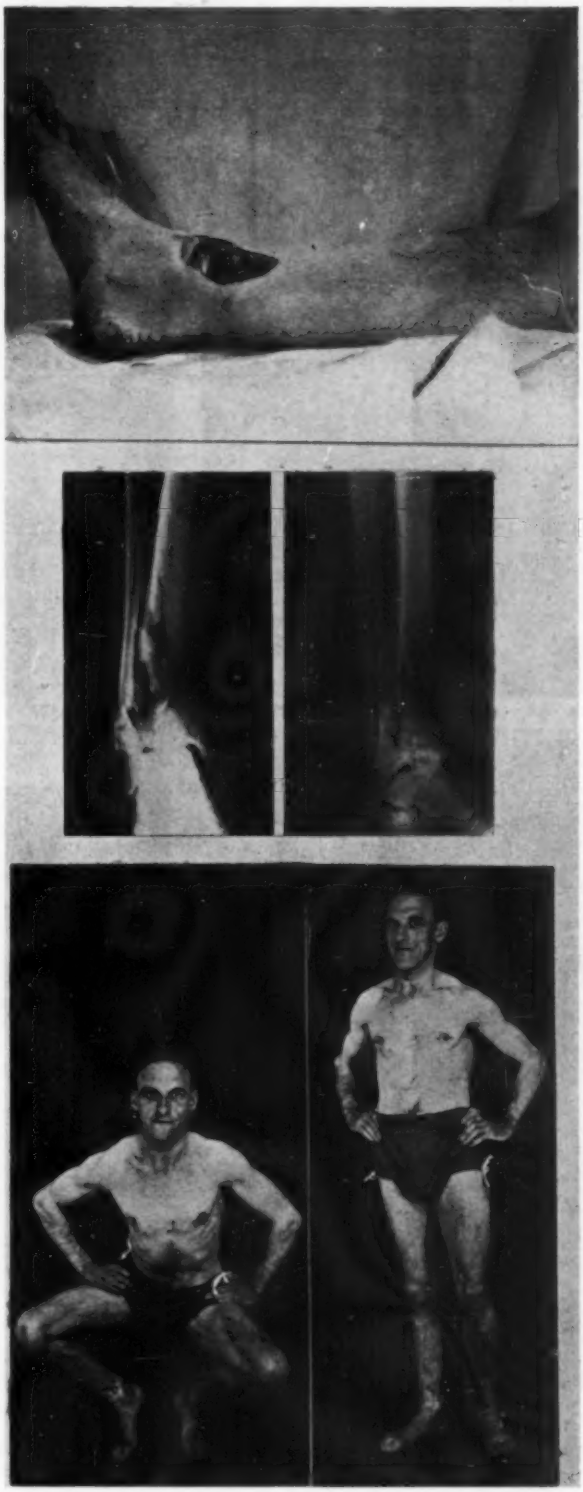

FIG. 359.—A, Photograph of inner aspect of wound 4 days from injury; B, Radiographs taken at the same time showing, in the lateral view, the gap between the upper and lower fragments of the tibia; C, Functional result 18 months later and after a sliding bone-graft had been inserted. Though the movements of the knee were not limited the ankle was stiff, causing pain on walking until 2 years after an arthrodesis which was performed elsewhere. (*From* Florey, 1952. By courtesy of the Oxford University Press.)

a position of equinovarus. A sliding bone-graft was not sufficient to maintain the foot in the normal position when walking out of plaster was started, and two later attempts at arthrodesis, at yearly intervals, were unsuccessful, the patient never being able to walk without pain. Amputation was performed consequently in the fifth year.

The second patient had the most severely injured limb in the series (*Fig.* 360 A). The tibia was fractured in 4 places, a gap of several inches being present in the mid third and there was also much loss of muscle and skin. The gap was still further widened when the opposing fragments separated as sequestra, so that a space of 5 inches was left to fill in. Nevertheless, each stage of plastic repair succeeded the previous one without mishap and, apart from the delay caused by successive sequestrectomies and an attempt to peg the tibial fragments to the fibula, the transfer of an 8-inch graft from the shaft of the opposite tibia followed uneventfully (*Fig.* 360 D). This graft apparently strengthened throughout the ensuing year and the patient was active in his walking plaster (*Fig.* 360 E) until he met with a motor accident in which the femur of the injured leg was fractured above the condyles. Some six weeks later sinuses appeared over the graft and at operation it was found lying in a bed of sterile pus. It was assumed that an injury sevère enough to fracture the femur above its condyles was also sufficient to sever the vascular connexions between tibial fragments and graft.

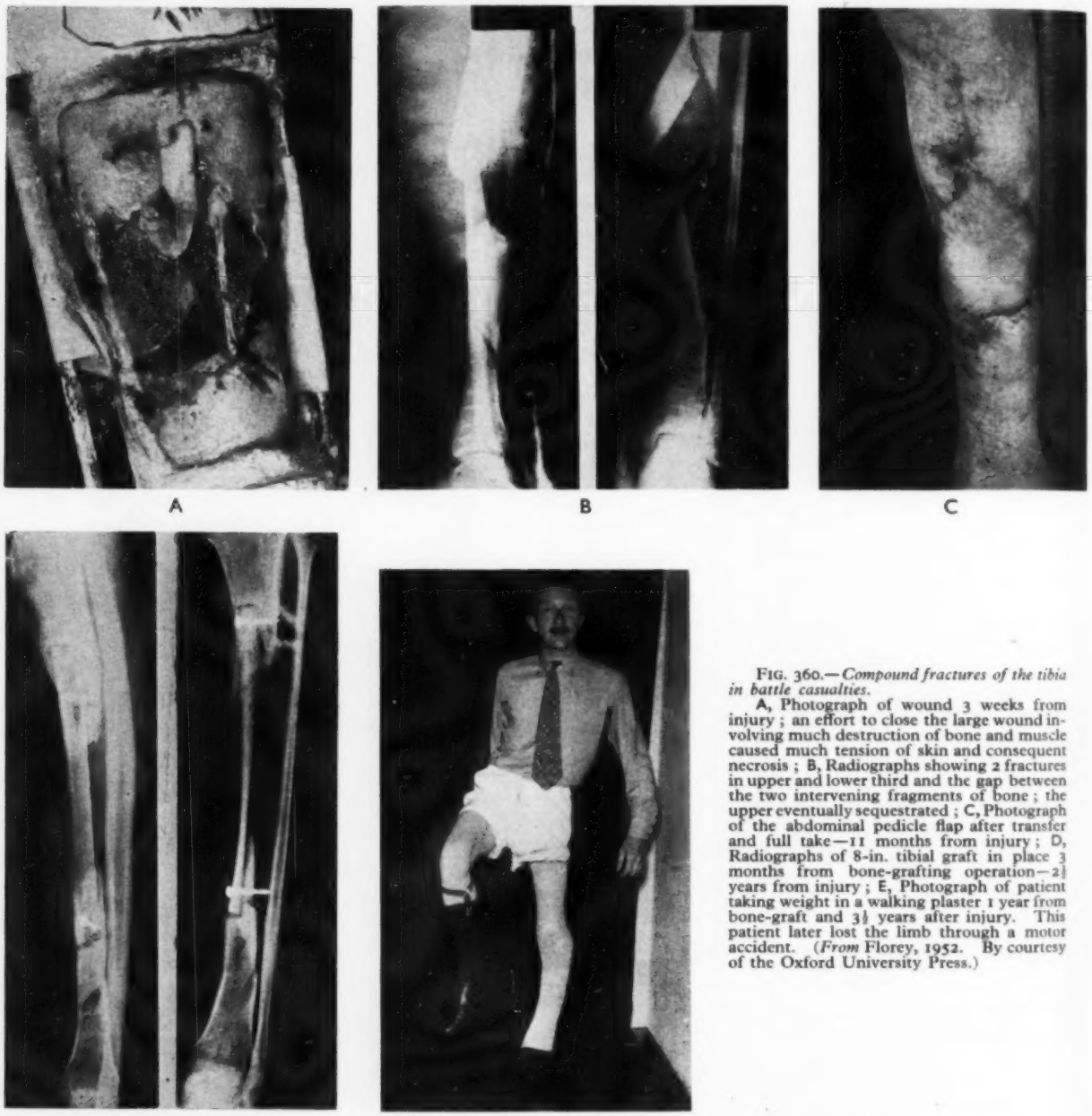

FIG. 360.—*Compound fractures of the tibia in battle casualties.*

A, Photograph of wound 3 weeks from injury ; an effort to close the large wound involving much destruction of bone and muscle caused much tension of skin and consequent necrosis ; B, Radiographs showing 2 fractures in upper and lower third and the gap between the two intervening fragments of bone ; the upper eventually sequestrated ; C, Photograph of the abdominal pedicle flap after transfer and full take—11 months from injury ; D, Radiographs of 8-in. tibial graft in place 3 months from bone-grafting operation—2½ years from injury ; E, Photograph of patient taking weight in a walking plaster 1 year from bone-graft and 3½ years after injury. This patient later lost the limb through a motor accident. (*From* Florey, 1952. By courtesy of the Oxford University Press.)

Amputation above the knee was advised and accepted by the patient.

The findings (of 40 compound fractures of the tibia) may be summarized as follows :—

5 involved the upper third
12 „ „ lower „
23 „ „ middle „

In 18 the fibula was also broken and in 6 a gap of 2 to 5 inches was present between the ends of the bony fragments.

Five to six years after wounding the results were as follows :—

Functional Results.—

10 were walking unaided within 6 months of injury
16 „ „ „ „ 1 year „
7 „ „ „ „ 18 months „
4 „ „ „ „ 2 years „
1 was „ „ „ 6 years „
2 had amputations after 5 years.

Full and free movement had been regained in 15; some limitation, mainly at the ankle, was present in 20; a stiff or ankylosed ankle was present in 4, and an ankylosed knee in 1.

Healing.—

28 had remained healed since discharge from the Unit.
12 had intermittently discharging small sinuses.
2 further operations had been performed elsewhere since discharge—curettage of bone in one and arthrodesis of the ankle to avoid pain in the other.

DISCUSSION

It was decided to make a long-term study regarding the effect of penicillin in battle casualties because short-term problems had already been dealt with (BRITISH JOURNAL OF SURGERY, 1944, **32**, Special Penicillin Issue). It had been demonstrated in North Africa and Italy that, with the aid of penicillin, the number of wounds harbouring pyogenic cocci was much reduced (Thomson, 1944; Bentley and Thomson, 1945); consequently early secondary suture of soft-tissue wounds could be undertaken with little risk and the maximum expectation of primary union (Bentley and Thomson, 1945; Brown, 1944 and 1945). There had been much reduction in the mortality from gas gangrene (Jeffrey and Thomson, 1944), from compound fractures of the femur (Furlong and Clark, 1944; McEwen and Bickerton, 1944), and from traumatic empyemata (d'Abreu, Litchfield, and Thomson, 1944).

With the institution of routine prophylactic injections of penicillin in the British Liberation Army from D Day onwards, the chances of development of pyogenic infection and of gas gangrene had been minimized (Porritt, Debenham, and Ross, 1945; Mitchell, 1947). Thus, though immediate risk to life and poor healing of soft-tissue wounds had been largely circumvented, it remained to be seen whether the sequelæ of chronic infection might also be prevented.

Compound fracture of bone caused by an explosive injury carries with it the menace of chronic infection, and consequent pain and limitation of movement in neighbouring joints. Of these, injuries to the tibia are among the most difficult to deal with owing to the poor blood-supply to the subcutaneous surface of the bone and the risk of limiting ankle movement, so important a factor in walking: sepsis here has serious consequences if not controlled. It was thought that, if satisfactory function could be restored to these limbs, it would be justifiable to retain others in the expectation of restoring them to use.

These results seem to justify the retention of limbs fractured in battle for the benefit of the men, who were all in their twenties or early thirties. An artificial limb is seldom considered as anything but a second-best substitute, but whether the country can afford so much time, skill, and labour as was devoted to their care is perhaps debatable. It is to be remembered, however, that none were continuously in hospital for more than the first 8 months, and while waiting for further operations were often able to occupy themselves usefully. Moreover, the disability pension paid by the Ministry of Pensions after 6 years to each man whose leg was saved varied between 0 and 40 per cent compared with 40 per cent to 80 per cent disability pension for the rest of his life had the leg been amputated.

It is not possible to assess accurately the part penicillin played in assisting repair. Clinical sepsis was not completely avoided if fever during the first 6 weeks and some suppuration are taken as an indication of its presence, but the pus was slimy and dull, not thick and creamy as was usually seen in septic wounds in pre-penicillin days, nor was the surrounding area of skin inflamed and œdematous. The extraordinarily rapid return to health of these men, with a minimum of help from blood transfusions, also indicated the power of penicillin to limit invasive infection.

It might be argued that the prevalence of penicillin-resistant staphylococci had not become a problem in the early days of repair of these wounds, but it still remains doubtful whether these organisms alone often play a major part in invasive rather than superficial sepsis (Sherris and Florey, 1951; de Pape and McEwen, 1951). They were not a serious problem in a Chronic Infection Unit for battle casualties of long standing up to the latter part of 1951, when the Unit was closed. It is possible that the passage of such organisms from one patient to another may increase both their resistance and their virulence. Other means of dealing with them, however, are now at hand so that the effect of controlling sepsis remains much the same.

However, the frequency with which penicillin-sensitive staphylococci appeared in wounds gave some indication of how easy it would have been for sepsis to arise and prejudice the results had not the regular bacteriological reports enabled these organisms to be detected and removed before they had had an opportunity to produce clinical signs. The almost complete absence of *Str. pyogenes* from their wounds while the men were in the ward was also another indication of the advancement of repair by the control of sepsis.

We wish to thank all those surgeons who lent their aid and skill to this enterprise, particularly Sir Max Page, who took care of the unit at a hazardous

tsage in its career, and Professor Pomfret Kilner for his sympathy and encouragement as well as for his help. The radiologists, Drs. F. H. Kemp, D. H. Jones, and R. Spalding Smith and their assistants, gave valuable advice on the progress of the fractured limbs, and without the help of the bacteriologists the work of the Unit would not have been possible.

To Sisters Ford, Mankellow, and A. Nicholson and their nursing staffs who successively worked for the men's benefit, much credit is due for the final results, as well as to the photographers who made them demonstrable.

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SHORT NOTES OF RARE OR OBSCURE CASES

A CASE OF LIPOMA OF THE BRONCHUS TREATED BY TRANSPLEURAL BRONCHOTOMY

BY E. GARSIDE BREWIN

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THE bronchus is a common site of neoplastic change, and the vast majority of tumours are malignant. Benign tumours are rare ; the 'adenoma' is most frequently encountered, simple connective-tissue tumours being excessively rare.

The present case is considered worthy of report, since only 12 examples of bronchial lipoma appear to have been reported previously (Langston, 1950 ; Carlisle, Leary, and McDonald, 1941) and since it was found possible to achieve local removal of the tumour by a non-mutilating procedure.

CASE REPORT

A man of 63 years was first seen on Aug. 22, 1950, complaining of cough, wheezy respiration, and dyspnœa. These symptoms had been present for about six weeks. He had little or no sputum, had never had hæmoptysis or pain in the chest.

ON EXAMINATION.—The patient was found to be rather barrel-chested and emphysematous with râles present over both lung fields. There was slight mediastinal shift to the left. These apart, there were no abnormal physical findings.

Radiologically.—Partial collapse of the left lower lobe was evident.

Bronchoscopy.—Cords, trachea, and carina normal. On entering the left main bronchus, about 1 cm. below the carina, a rounded pale pink, cherry-like tumour could be seen, moving up and down on respiration as if having a pedunculated attachment ; it almost filled the left main bronchus. The actual site of origin of the tumour could not be seen ; the mucosa over it was quite smooth and the whole appearance was that of a simple tumour.

It was considered at this time that the diagnosis was probably adenoma. A biopsy was taken, it not being possible to pull out the tumour bodily.

Biopsy Report.—No lesion is demonstrated in the biopsy from the bronchus. The fragment of mucous membrane is covered with tall, columnar, ciliated epithelial cells ; numbers of mucous glands and muscle-fibres are also present.

In view of the rather unhelpful biopsy report, it was thought advisable to proceed to thoracotomy and explore the tumour by bronchotomy, with a view to local removal should it be practicable and pathologically desirable.

AT OPERATION (Aug. 31, 1950).—With the patient in the prone position, left thoracotomy was carried out through the bed of the sixth rib. The left main bronchus was cleared and an incision made longitudinally in the non-cartilaginous portion, between traction sutures. The tumour was easily delivered from the bronchus and was found to be attached by a slender stalk at a point just above the upper lobe orifice on the lateral wall ; it was the size of a small cherry. There was no other abnormality of the bronchus. The stalk was detached from the mucosa and the base lightly cauterized with silver nitrate solution. The incision in the bronchus was closed with interrupted thread sutures, the peribronchial connective tissues and pleura being approximated over the suture line. The thoracotomy wound was then closed.

The patient made a good recovery and when last seen, fifteen months after operation, was quite well and symptom-free.

PATHOLOGY.—The tumour has been examined by the panel of pathologists of the Benign Tumour Registry of the Society of Thoracic Surgeons. By five members of this panel it was considered to be a lipoma, and the sixth regarded it more as a hamartoma composed mainly of fat, but including mucus-secreting glands.

My thanks are due to Mr. Gordon Cruickshank, in whose wards this patient was treated.

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PENETRATING INJURY OF THE ABDOMEN IN A BOY AGED 16 YEARS

By R. SPENCER, Liverpool

The following case history is sufficiently interesting to merit recording both on account of the unusual circumstances and the type of injury involved.

CASE REPORT

The patient, N. P., aged 16 years, was one of nine casualties, all children, admitted as a result of an explosion in a street on the evening of Oct. 28, 1951. It was later ascertained that the cause of the explosion was due to a ship's distress signal which had been stolen from a barge moored on the bank of the River Mersey.

The patient was suffering from acute abdominal pain and was severely shocked. There were multiple puncture wounds on both legs and a few on the lower abdomen, but these appeared to be superficial. There was a larger puncture wound three-quarters of an inch in length situated in the midline of the epigastrium from which bubbles of gas were seen to be escaping. There was generalized abdominal rigidity and tenderness. A diagnosis of perforation of an intra-abdominal viscus was made, probably of the stomach. The blood-pressure was 130/70 and the pulse-rate 86 per minute. Morphine was given and blood transfusion was commenced.

At Operation.—He was taken to the operating theatre two hours after admission and the abdomen was explored under a general anæsthetic of gas, oxygen, and ether.

The peritoneal cavity contained a large quantity of pure bile and some blood. This was sucked out before examining the intraperitoneal viscera. There was a large stellate laceration of the right lobe of the liver, a perforation of the under-surface of the gall-bladder, about three-quarters of an inch in length, from which bile was escaping, and a small irregular laceration of the anterior surface of the first portion of the duodenum, also about three-quarters of an inch long, from which duodenal contents and gas were escaping. The interior of the duodenum was exposed by extending the laceration in the line of the gut and the margins of the duodenum were held apart with Allis's forceps. There was a tear of the posterior wall of the duodenum corresponding with the anterior perforation. (*Fig.* 361.) A probe passed into this laceration came up against a metallic foreign body lying in the cellular tissue immediately behind the duodenum. It was removed by taking hold of it in artery forceps. The tear of the posterior wall of the duodenum was repaired by three or four interrupted catgut sutures (No. 00). The anterior wall of the duodenum was repaired in the manner of a pyloroplasty operation in order to avoid narrowing of the lumen. Two layers of catgut sutures were used, the first continuous and the second interrupted. The great omentum was stitched over the suture line as an added precaution against leakage. The perforation of the gall-bladder was closed by means of a purse-string stitch. The liver was repaired in the classical manner by means of deep sutures through the substance of the liver and stitches at right-angles to these and outside them crossing the area of damage. Hæmostasis having been secured, the abdomen was closed in the normal way leaving a drain and a length of gauze draining Hartmann's pouch.

Post-operative Progress.—Blood transfusion was continued—two litres altogether were given, and this was followed by intravenous glucose saline (*N*/5 saline and 5 per cent dextrose). He received approximately three litres of fluid intravenously in each twenty-four hours. The administration of intravenous fluid was continued for three days. The patient was pyrexial for about ten days and the wound discharged large quantities of blood-stained bile. The stomach was kept empty by continuous aspiration. He was given A.T.S., distaquaine 1 c.c. daily, and streptomycin 1·5 c.c. twice daily for ten days. The gauze and the tube were removed on the third post-operative day. A modified Sippy diet was commenced on the fifth day. He was given an enema on the fifth day with good result. His bowels began to move normally at the end of ten days. The sutures were removed on the tenth post-operative day. The wound continued to discharge bile for another ten days following this, but this became negligible at the end of three weeks from the time of his operation and finally ceased. A cholecystogram carried out before his discharge failed to outline his gall-bladder. A barium meal and radiographs showed there to be normal emptying of the stomach without narrowing of the duodenum. He was discharged on Dec. 12, 1951.

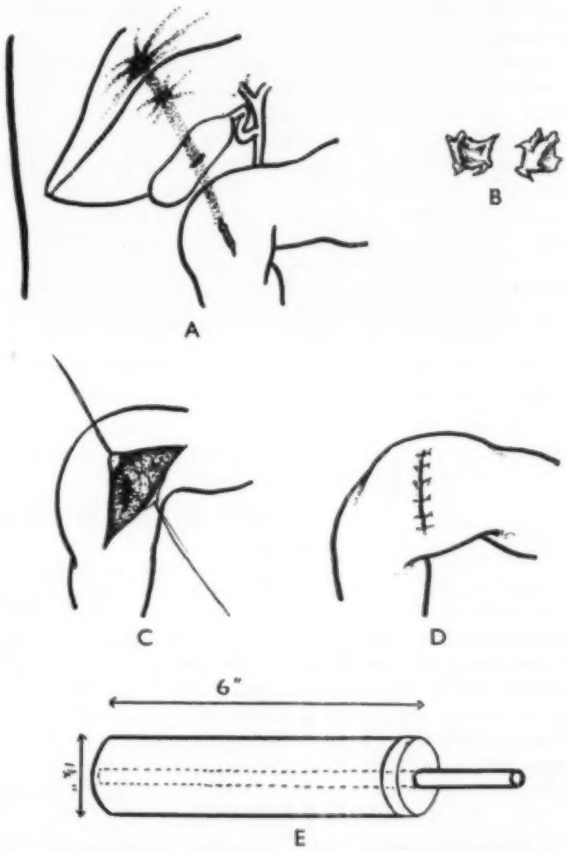

Fig. 361.—Diagram to illustrate penetrating wound of the abdomen: A, Track of metallic foreign body; B, Foreign body (half size); C, Wound in posterior wall of duodenum; D, Repair of anterior wall of duodenum; E, Distress signal which caused the explosion.

COMMENT

This type of injury is rare in civilian surgery but is common enough in air-raid and battle casualties. With the help of the C.I.D., information was obtained concerning the cause of the explosion. This was due

to a rocket distress signal. It consists of a cylindrical metal container about six inches in length and an inch in diameter full of explosive powder and closed at the top. Running through the centre of the cylinder and attached at its base is a metal rod which projects through the cap and is fitted with some kind of rocket arrangement. The rocket itself had been lost or removed. This type of distress signal is now obsolete and has been replaced by a more modern and less dangerous type. Several boys had obtained one of these distress signals and having removed the cap had applied a match to the powder. The only portions of the signal which remained were embedded in the victims. Fortunately most of the injuries were superficial and subcutaneous and intramuscular. One small boy suffered severe damage to one eye requiring enucleation. (*Fig.* 361, B, E.)

Injury to the posterior wall of the duodenum is of particular importance for three reasons: First, it may easily be missed; secondly, the difficulty of access; thirdly, the proximity of other very important structures, e.g., the inferior vena cava, the aorta, the common bile-duct, and the pancreas. In one previous case a patient who was pushing a ladder along a road on a hand-cart collided with a stationary car and received a sudden unexpected blow in the epigastrium. Clinically he had all the signs of a ruptured intraperitoneal viscus. At operation, in spite of a prolonged search, no obvious lesion was detected although there was blood-stained fluid in the peritoneal cavity. This patient died forty-eight hours later and at autopsy was discovered to have a severe cellulitis of the retroperitoneal tissues resulting from a laceration of the distal fixed portion of the first part of the duodenum.

There are two obvious ways of gaining access to the posterior surface of the duodenum. The first is to incise the peritoneum on the lateral aspect of the descending portion and to turn the duodenum over towards the left. This has obvious drawbacks where one may be dealing with multiple injuries and traumatized tissues. The area is normally very vascular and the tissues are friable when injury has occurred. The second is to approach the posterior wall by a transduodenal route. It is easy to make this decision where there is already an incision of the anterior wall but it might have to be decided on without this guide if there is reasonable suspicion of hidden damage.

SUMMARY

A case of penetrating wound of the abdomen involving laceration of the liver, gall-bladder, and both walls of the duodenum has been described. The method of dealing with a damaged posterior wall of the duodenum has been discussed.

I should like to thank Dr. Crosbie, Medical Superintendent, for permission to publish this account and also Miss Barbara Duckworth for her excellent diagrams.

NEURILEMMOMA OF THE TONGUE

By A. St. CLAIR ROBERTSON

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Neurilemmoma (Syn. Neurinoma, Schwannoma) of the tongue is an uncommon condition. Gnassi and Borrone (1938) in a review of the literature collected 9 such cases and added another. Further cases added by Ricard (1939), Willis (1948), and Hung (1949) have brought the total to 13 cases.

CASE REPORT

A married woman, aged 28 years, complained of a lump on her tongue. Fourteen years previously a red spot had appeared at the tip of her tongue and this had gradually increased in size. It was entirely symptomless until recently, when she found she was unable to wear her dentures because of the enlargement of the tip of her tongue, and her speech was slightly slurred.

On Examination.—A swelling, 4 cm. in diameter, was seen occupying the tip of the tongue. The mucous membrane over it was normal and was not adherent to the tumour. The tumour was firm but not hard; there was no tenderness. No enlarged cervical glands were to be found (*Fig.* 362).

At Operation.—Under general anæsthesia, a transverse incision was made proximal to the tumour and the tumour and the tip of the tongue were excised. The defect was closed by direct suture of the margins of the wound.

Pathology.—

Macroscopical Appearance (*Fig.* 363).—This specimen consisted of the distal extremity of the tongue and measured 3·5 × 2 × 2 cm. It was rounded and ovoid except for the junction with the tongue. The cut surface (transverse) showed a well demarcated mass 3 × 2 × 2 cm. in size, with a thin apparent capsule 0·1 cm. thick, greyish in colour, with a central cavity measuring 2 × 1 × 1 cm. which contained recent blood.

Microscopical Appearance.—

1. *Hæmatoxylin and Eosin.* This showed cells arranged in an orderly manner amongst fibres; in other parts the fibres were more dense, the cells more elongated and arranged in interlacing wavy lines which showed regimentation or a palisaded pattern.

2. *Van Gieson.* This showed little fibrous tissue apart from the capsule of the tumour. This is a neurilemmoma. It is apparently benign.

Post-operative Course.—This was uneventful except that for a few days after the operation there was considerable swelling of the tongue. This quickly subsided. The tongue was completely healed when seen a month after operation. There was no residual speech defect.

COMMENT

In a review of the literature available on this condition it was found that there was uniformity of symptoms and signs: a slow growing symptomless tumour which made the patient seek advice, only because of its size or interference with speech or swallowing.

At operation in the majority of cases it was found that the tumour was enucleated with the greatest ease, an almost bloodless procedure. If a correct diagnosis, pre-operatively, had been made in this case a similar procedure would have been possible.

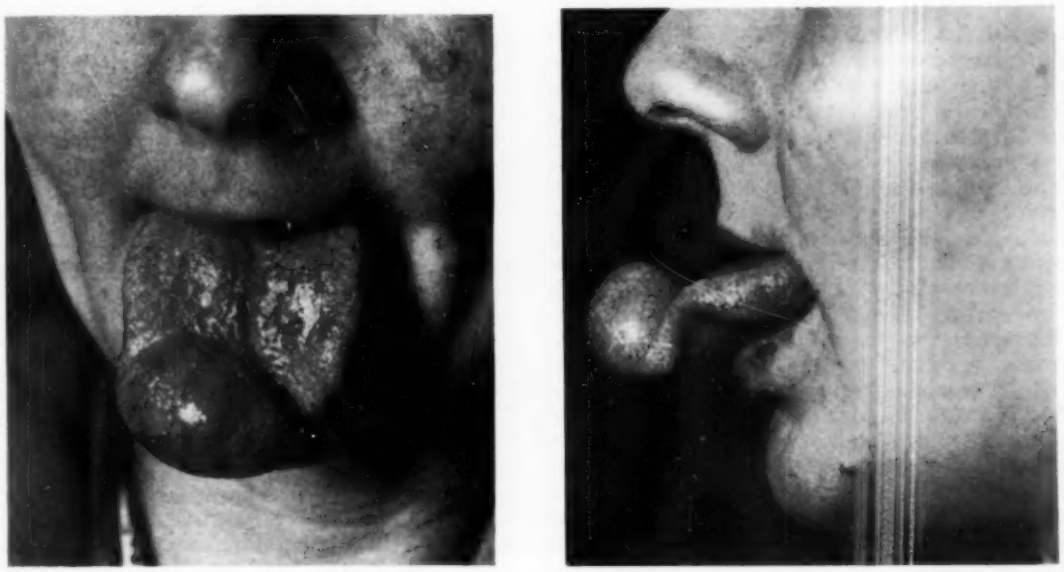

FIG. 362.—Photographs to show the appearance of the tumour in relation to the tongue.

FIG. 363.—Photograph of the cut surface of the specimen to show the capsule tumour and large cystic space filled with recent blood-clot. (× 2.)

Recurrence of a neurilemmoma of the tongue has not been recorded, but it must be noted that recurrence of a neurilemmoma has been recorded in cases in which it was situated in the floor of the mouth.

From 1941 to 1951, 30 specimens of neurilemmoma have been examined histologically in the Pathological Department of this hospital (11 biopsy specimens only). In 7 cases there was evidence of a central area of degeneration, degenerative changes, or a cystic space or spaces, although none as large as that found in this case.

SUMMARY

A case of neurilemmoma of the tongue is described. It is suggested that in the differential diagnosis of swellings of the tongue this condition should be considered, particularly because of the ease with which it may be treated.

I should like to thank Mr. J. C. Anderson, in whose ward this case was treated, for permission to publish this case; Dr. J. L. Edwards for his advice on the pathology and for allowing me to use his records; and to Mr. J. F. V. Larway for the photographs.

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ENTEROGENOUS CYST OF THE STOMACH

REPORT OF A CASE

By A. LYALL, Greenock

According to Ladd and Gross (1947) duplications of the alimentary tract may be found at any level from the base of the tongue to the anus, although it is commonest in the small intestine. They emphasize that there are three features common to all of them. They are in close proximity to some part of the alimentary tube; there is a smooth muscle coat, frequently in two layers; and the lining epithelium is similar to that of some part of the stomach, small intestine, or colon. The duplication may be connected with the lumen of the adjacent alimentary tract or be segregated from it to form a cyst. According to Donovan and Santulli (1947) the condition occurs most frequently in the lower ileum, cysts of enteric origin being differentiated from those arising from Meckel's diverticulum by their situation on the mesenteric aspect of the bowel. Mayer (1935), in mentioning the rarity of enterogenous cysts, pointed out that he had failed to find a single example in a review of 80,000 post-mortem examinations. Kettle (1933) collected 88 cases of which 71 were found in the small intestine and 17 in the colon. They have also been reported in the duodenum (Booher and Pack, 1946; Pachman, 1939), in the mediastinum (Valle and White, 1946), and the base of the tongue (Sherwin, 1938). These cysts are very rare in the upper part of the alimentary tract, and according to Orgias (1943) enterogenous cyst of the duodenum has been described on only ten occasions, four of the patients surviving operation.

A study of the literature shows that up to the present only 8 enterogenous cysts of the stomach have been described. Lyons (1916) described a cyst lying along the greater curvature in a 19-day-old baby. Ladd and Gross (1940) described two cases, one in an infant of two months and the other in a child of nine years. Cases were also reported by Ferrara (1942) and Cabot (1928), the latter finding the cyst as a 'tumour' the size of a plum in the stomach of a 7-year-old child. In Wendel's case (1911) the cyst lay on the greater curvature of the antrum of the stomach in a 30-year-old woman. In Miller and Ginsberg's case (1950), the cyst lay behind the pylorus extending from the anterior to the posterior wall of the stomach so that the pyloric canal was constricted from below. McNeer and Spitz (1951) reported a case in a male of 34 who had a cyst occupying the distal third of the stomach.

The enterogenous cyst has a wall of smooth muscle and an inner lining of mucosa. The epithelium may not be the same as that of the part of the alimentary tract to which it is attached. Thus Ladd and Gross (1947) mention that a cyst of the tongue was found to be lined by colonic mucosa and a cyst of the rectum partially by gastric epithelium. The content is usually a thin glairy, mucoid fluid. According to McNeer and Spitz (1951) enterogenous cyst of the stomach is generally recognized early in life, due to the production of obstructive symptoms. In Cabot's case (1928), the child came under medical care because of 'cyclical vomiting'. In the older age group, there may be merely a sense of fullness after meals or the symptoms may be those of peptic ulceration, even with hæmatemesis.

In view of the extreme rarity of enterogenous cyst of the stomach, the following case is worth recording.

CASE REPORT

History.—J. McN., aged 9 years, was admitted to hospital on Feb. 18, 1950, with a history of recurring vomiting over a period of some months. This had been accompanied by a marked loss of weight and the provisional diagnosis of tuberculous disease of the abdomen had been made. Three days before admission, vomiting had started and had continued so that he had become markedly dehydrated. This had been accompanied by upper abdominal pain.

On admission, the child was very dehydrated and examination showed a tender mass in the epigastric region. A provisional diagnosis of intussusception was made and operation carried out soon after admission, the dehydration being corrected by intravenous therapy.

At Operation.—The abdomen was opened by a right upper rectus-splitting incision. The distal half of the stomach was replaced by a firm mass measuring 9 × 6 cm. It was thought to be a solid tumour occupying the pyloric half of the stomach. A partial gastrectomy was carried out by the Polya method, about two-thirds of the stomach being removed, and the jejunum being brought up for anastomosis through the transverse mesocolon, the afferent loop being attached to the lesser curvature of the stomach.

The boy made an uninterrupted recovery and was discharged two weeks later. A barium examination a few weeks later showed that the stomach was emptying well. In the following months the boy made good progress and rapidly increased in weight. Since the operation he has remained well.

Pathology.—Examination of the resected specimen showed that it consisted of the pyloric end of the stomach with a large cystic mass growing into it from above. Section showed that the mass consisted of two cysts connected by an opening measuring 7 mm. in diameter, the larger being proximal and measuring 6 × 4·5 cm., and the smaller bulging through the pylorus into the duodenum and measuring 2·5 × 2·5 cm. (*Fig.* 364.) The pyloric half of the stomach was reduced to a narrow slit lying along the inferior aspect of the mass and it was difficult to understand how any nourishment could have passed along it for some time. The content of the cyst was a clear glairy fluid of neutral reaction. The wall of the larger cyst was white in colour; the smaller one appeared hæmorrhagic, but this was considered to be due to trauma at the operation. There was no connexion with the cavity of the stomach or duodenum. There was a recognizable muscle layer which was continuous with that of the adjacent upper part of the stomach.

Histology.—The wall of the cysts was lined by columnar epithelium, the cytoplasm being clear with elongated nuclei which were basophilic and lay at the base of the cell. (*Fig.* 365.) No goblet cells were seen. The surface was flat with no evidence of valvulæ conniventes, but there were slight depressions at places representing the mouths of glands which lay in the fibrous stroma of the mucosa. These glands appeared to be in different stages of activity. Thus, in some, the lining cells of the acini were cubical

with neutrophilic and rounded or elongated nuclei lying in the basal part of the cell. In others, the cells were more flattened, the cytoplasm being clear and only faintly stained. There were very occasional groups of lymphocytes in the mucosa just superficial to the muscularis mucosa which was present as a very definite layer of muscle-fibres rather loosely arranged. The submucosa consisted of a layer of loose fibrous tissue carrying prominent blood-vessels and spreading into the muscular layer at places. This muscular coat was very prominent and consisted of both longitudinal and circular fibres.

DISCUSSION

This case presents points of interest. It appears to be the ninth case of enterogenous cyst of the stomach to be described. The cyst was closely attached to the stomach and, in fact, it appeared to be incorporated in the gastric wall and separation would obviously have been impossible. Ladd and Gross (1947) stressed this close union and pointed out that although there may be a slight furrow between the duplication and the intestine, attempts to dissect the cyst away would verify the absence of any plane of cleavage. Wooler (1950) also mentions the difficulty of separating these cysts from the adjacent bowel and adds that this is a means of distinguishing duplications of the intestinal tract from mesenteric cysts. Donovan and Santulli (1947) mention that duplications always share the same blood-supply as the contiguous bowel and that this is of great importance if resection has to be performed. In the present case, partial gastrectomy was the only possible treatment and was followed by a dramatic improvement in the child's health.

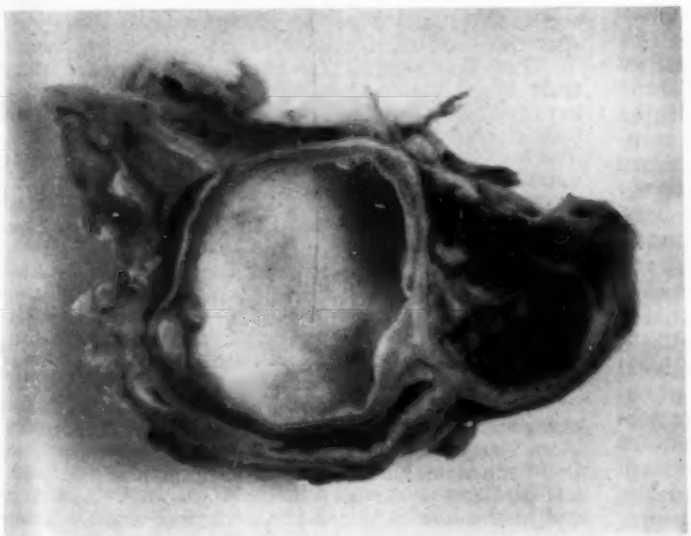

FIG. 364.—Section of cystic mass seen from posterior aspect. The larger cyst lies on the left and communicates with the smaller on the right by a small opening. The cavity of the stomach has been reduced to a narrow slit at the lower end of the specimen. The muscular coat of the cyst is continuous with the muscle layer of the gastric wall.

There have been several theories put forward to explain the origin of the enterogenous cyst. Lewis and Thyng (1907) in a series of dissections of the gastro-intestinal tract in lower animals and human embryos, frequently found nodules of epithelial cells which tended to become vacuolated and break off from the alimentary tract or remain in continuity to form a diverticulum. These tiny diverticula normally regress but it can be readily understood how one may remain to form an enterogenous cyst. Bremer (1944) suggested that there is actually an active growth of an epithelial bud from the intestinal mucosa into the loose submucosa where it forms the cyst which then burrows between the muscle-fibres. Johnson (1910) suggested that a defect in canalization might be a possible aetiological factor. In the six-weeks embryo, the lumen of the developing bowel is occluded by proliferating epithelium. The cells then secrete a fluid which is collected into vacuoles which are arranged in rows and run together to form a continuous lumen. He believed that in some cases a vacuole may persist to form a duplication. Ladd and Gross (1940) considered that the cysts were simply due to the pinching off of diverticula of the fœtal intestinal tract.

FIG. 365.—Low-power section of cyst wall. The lining cells are columnar epithelial in type. The muscularis mucosæ is well marked and there are numerous glands in the submucosa.

Most of the above views as to aetiology sound plausible, but a study of the cyst described in the present article and its incorporation in the wall of the stomach suggests a possible origin from a 'pinched off' diverticulum or a group of sequestrated cells. The formation of intestinal epithelium in the cyst points to this having occurred at a very early stage of the formation of the alimentary tract and makes its possible origin from a sequestration of cells appear to be the more likely one.

SUMMARY

A case of enterogenous cyst in a boy of nine years is described. He had suffered from cyclical vomiting for some months. Partial gastrectomy was successfully carried out. The aetiology of the condition is discussed.

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ISCHÆMIC NECROSIS OF A FLEXOR TENDON

BY A. J. DREW, WALSALL

NECROSIS of tendons is a common sequela of infection or trauma at certain sites. Both tuberculous and pyogenic infections may affect the synovial sheaths and necrosis of the tendon may ensue, but neither process should be difficult to diagnose on clinical grounds with microscopical aid. Little has been added to our knowledge of tuberculous tenosynovitis of the hand since Kanavel's (1923) excellent paper on the subject. He stressed the gross nature of the pathological changes and pointed out that the histology is similar to that of tuberculosis elsewhere in the body, being characterized by endothelioid-cell proliferation, round-cell infiltration, and giant cells. In 6 of his 14 cases there was fragmentation of the tendon involved.

Traumatic rupture and injury have been described and discussed at length by Mason (1930) and McMaster (1933), and there has been little advance in our understanding of the mechanism or pathology of the condition since. Mason demonstrated the strength of the normal tendon by reference to the case of a sailor in whom, as a result of trauma, the whole muscle belly was avulsed with the finger-tip. He gave a classification of tendon ruptures, and reported two cases with a traumatic rupture of a flexor tendon of the hand. McMaster concluded that, although direct trauma in which the tendon is caught between the striking object and underlying bone may occasionally cause a tendon to rupture, indirect trauma will not rupture a normal tendon. In experimental muscle–tendon–bone preparations from rabbits he confirmed his clinical conclusion that usually the tendon insertion, muscle origin, muscle belly, or musculotendinous junction ruptures and not the tendon. Considerable damage to a tendon is needed before rupture occurs. Double ligation causing ischæmic necrosis produces this effect only after an interval of seven weeks. These conclusions are borne out by the common clinical findings. Mallet finger and a tear of the patellar ligament occur at the bony insertion. Ruptures of the supraspinatus tendon, biceps brachii tendon, and extensor and flexor pollicis longus tendons are associated with osteo-arthritic changes in the joint. Tears of the hamstrings, quadriceps, and adductors of the thigh are in the muscle belly or at the musculo-tendinous junction.

These papers agree with clinical experience that the cause of traumatic rupture is usually apparent.

The following case does not seem to conform with any of the conditions already discussed, and from an extensive search of the literature it would appear not to have been described hitherto. The pathological findings are so unusual that it is felt that the case is worthy of being recorded.

CASE REPORT

HISTORY.—E. H., a married woman and a housewife, aged 31 years, attended hospital complaining of inability to extend completely the ring finger of her right hand for a month. Though she had noticed the disability, she had no clear recollection of its onset. A week later an aching pain started at the base of the fourth finger and radiated to the rest of the palm, and a few days after this she noticed a lump in the palm of her hand.

There was nothing of significance in her past history apart from attacks of epilepsy since childhood, said to have followed a head injury. These attacks were successfully controlled by a daily dose of epanutin and luminal. The last attack had occurred two months ago. On being questioned she said she thought that she usually fell on to her right hand. Apart from this, she had no recollection of injury or infection and otherwise she was perfectly well.

ON EXAMINATION.—The ring finger of the right hand was held in 20° of flexion at the proximal interphalangeal joint and almost full extension at the distal interphalangeal joint. There was no active flexion at the distal interphalangeal joint, but extension was normal. Active flexion at the proximal interphalangeal joint was full, but active extension was limited by 20°. Active movements at the metacarpophalangeal joint were normal except for slight limitation of extension. There was a firm, smooth, well-defined swelling, ¼ in. in diameter, in the palm, just proximal to the head of the fourth metacarpal bone and in the line of the tendons. It was slightly tender on pressure and moved proximally on flexing the ring finger. The rest of the hand was normal.

The condition was considered to be an unusual type of trigger finger in which the profundus tendon had become locked in the sheath, and operation was advised.

AT OPERATION.—Under a general anæsthetic, with the arm ensanguinated by a pressure cuff, a transverse incision in the distal palmar skin crease was made. The tendon-sheath of the ring finger was exposed. The flexor digitorum sublimus tendon appeared perfectly normal, but there was no sign of the proximal end of the flexor digitorum profundus tendon. The palpable lump was seen to comprise a white nodule lying in the proximal end of the digital synovial sheath, which was thin, translucent, glistening, and macroscopically normal. On opening the sheath the nodule was easily lifted out and had no attachments. It was then realized that it consisted

of about 3 in. of the flexor profundus tendon rolled up in a ball. When unrolled it had the appearance of dead tendon, was a dull matt white, completely avascular, with frayed ends (*Fig.* 366). The distal end of the tendon could not be found and there appeared to be no local abnormality to account for these peculiar findings.

PROGRESS.—The patient made an uneventful recovery and the wound healed by first intention. The final disability, as expected, was a complete absence of

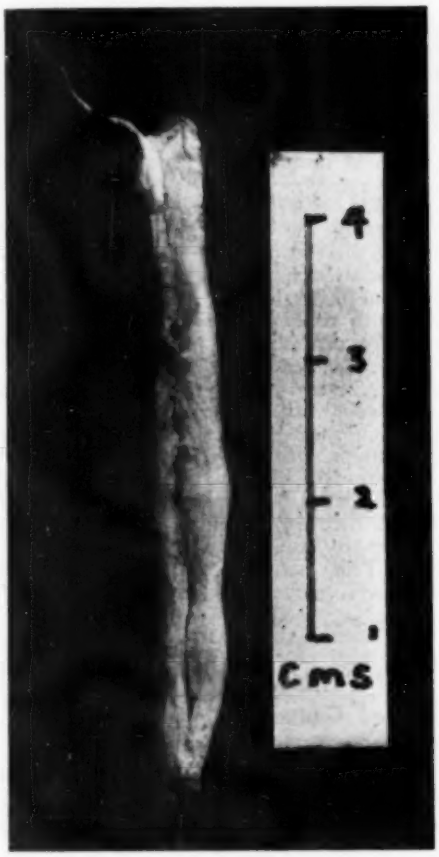

FIG. 366.—Specimen of ischæmic tendon removed at operation.

active flexion of the terminal phalanx, although full passive flexion remained. Full movements returned to the metacarpophalangeal and proximal interphalangeal joints, and full extension to the terminal interphalangeal joint.

THE SPECIMEN.—Dr. Navasquez examined the specimen and reported that microscopically, "the tendon shows ischæmic necrosis of 'coagulative' type with retention of gross histological structure. No inflammatory disease detected".

COMMENT

The unusual features of this case are the remarkable pathological findings and their aetiology. Had there been gross or microscopical evidence of infection, whether tuberculous or otherwise, a description of this case would not have been warranted. Again, though rupture of a flexor tendon is unusual, a single break might have been accepted as being due to trauma. The double break with a loose segment of tendon and the absence of inflammatory reaction needs further consideration and is not easy of explanation.

Edwards (1946) gives a complete and detailed description of tendon anatomy and points out that tendon is virtually dead, needing only a blood-supply to the tendon cells and moisture for lubrication. Even so, the vascular system is complete, although simple, consisting of a series of longitudinal arterioles in the interfascicular tissue or endotenon connected by numerous transverse anastomotic vessels. With the flexor tendons in their synovial sheaths this internal system only connects with the general vascular system at the musculotendinous and osteotendinous junctions and by way of the vinculæ. Because of these minimal nutritional demands it appears avascular, is destroyed, and sequestrates, or heals extremely slowly.

Howard (1941) agrees that the collagen fibres of the tendon fasciculi are inert and do not participate in metabolic processes, and that they are incapable of responding to irritation or trauma, except that of mechanical fraying or rupture. Severed tendons heal by granulation tissue and scar formation. This granulation tissue probably arises from adjacent structures and explains the tendency to adhesion formation and occasionally the erosion of a tendon by encroaching granulation tissue. He also suggests that single or repeated tendon injury may call forth diseased states which become chronic, and, at first glance, seem unrelated to the initial injury.

In this case an interruption of the blood-supply or ischæmic necrosis, of the tendon in that part most dependent upon its internal arterioles, is the most likely explanation of the pathology. The severance of the necrosed section at both ends accords with the pathological fact that separation occurs at the junction of living and dead tissue whether skin, bone, or tendon, and is explained by the reaction of inflammation at this site. The cause of the ischæmic necrosis would at first appear to be the simple one of single or repeated trauma during her epileptic attacks. Single or repeated blows to the palm of the hand are of everyday occurrence in modern industry, and although the bones may suffer as with the pneumatic driller, the soft tissues seem well able to protect themselves, as is shown by the absence of similar cases.

Apart from recording the unusual features of this case, it is hoped that it may call forth reports of cases of a similar nature, for only in this way will the true aetiology become apparent.

It is a pleasure to acknowledge the help and encouragement given by Mr. Grant Massie in the preparation of this paper.

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REVIEWS AND NOTICES OF BOOKS

The Science and Practice of Surgery. By W. H. C. ROMANIS, M.A., M.B., M.Chir. (Cantab.), F.R.C.S. (Eng.), F.R.S. (Edin.), Senior Surgeon and Lecturer on Surgery, St. Thomas's Hospital; etc.; and PHILIP H. MITCHINER, C.B., C.B.E., T.D., M.D., M.S. (Lond.), F.R.C.S. (Eng.), Hon. Surgeon to H.M. The Queen; Vice-President, Royal College of Surgeons of England; Consulting Surgeon, St. Thomas's Hospital. Ninth edition. In two volumes. 6 × 9¼ in. Vol. I, General Surgery, pp. 872 + viii, with 402 illustrations and 20 plates. Vol. II, Regional Surgery, pp. 1019 + viii, with 326 illustrations and 8 plates. 1952. London: J. & A. Churchill Ltd. Vol. I, 32s.; Vol. II, 36s.

THE new ninth edition of this two-volume work on surgery has been thoroughly revised by the authors. It is sad to think that just as this new edition appeared we heard of the sudden death of one of the authors, Mr. Philip Mitchiner. He was a sound practical surgeon with a great flair for teaching and he will be greatly missed by surgeons both at home and abroad.

The fact that this work first appeared some twenty-five years ago and has passed through nine editions speaks for itself and proves its usefulness and popularity.

The new edition still maintains its place as an excellent text-book for the student, for in it he will find all that is required for his final examinations in surgery. For the practitioner it will form a reference guide for all cases of difficulty which may arise in his practice. We have only one fault to find with the new edition and that is that some of the illustrations which date back to the earlier editions are reproduced badly and are not distinct; we hope this will be remedied in the next edition.

The Knee and Related Structures. Injuries—Deformities—Diseases—Disabilities. By PHILIP LEWIN, M.D., F.A.C.S., F.I.C.S., Professor and Chairman of Department of Bone and Joint Surgery, Northwestern University Medical School; etc. 6 × 9¼ in. Pp. 914, with 333 illustrations and 2 coloured plates. 1952. London: Henry Kimpton. 120s.

THIS large volume of nearly a thousand pages deals comprehensively with all conditions and diseases of the knee-joint. Professor Lewin is to be congratulated on his great task of bringing all available knowledge concerning the knee-joint into one book. After considering the anatomy and physiology of the knee-joint, the author deals in detail with the various aetiological factors in knee-joint disturbances, including the internal derangements of the joint.

Fractures in and around the knee are well described and their treatment discussed in a very practical way.

As Professor Lewin served in both world wars it is not surprising to find that his various methods of treatment of knee-joint injuries are sound and practical. It is interesting to read of Gordon-Taylor's ten commandments which should be used as a guide by those who have to undertake a primary or emergency amputation, and less interesting to find no reference whatever to Gordon-Taylor in the extensive bibliography at the end of the book.

This book is extensively illustrated with good pictures on the whole, yet the coloured plates are not very good, for there is not enough contrast in the pathological details. The book will undoubtedly become the standard reference volume on the knee-joint for many years to come.

Surgery of the Œsophagus. By R. H. FRANKLIN, M.B., B.S., F.R.C.S., Senior Lecturer and Surgeon, Post-graduate Medical School of London. 5½ × 8½ in. Pp. 222 + xii, with 86 illustrations and 4 plates. 1952. London: Edward Arnold & Co. 42s.

THIS is an excellent monograph dealing with the surgery of the gullet; a very neglected branch of surgery in the past, yet which to-day may be said to be coming into its own. It is to pioneer surgeons such as Trotter and Grey Turner who have led the way for a better understanding in the treatment of malignant disease of the œsophagus that we owe so much.

Mr. Franklin, who has devoted much time and thought to the surgery of the œsophagus both in children and adults, is just the right person to write this monograph. His long association with Grey Turner at Hammersmith and their combined efforts to find out the best methods of dealing with cancer of the œsophagus have indeed borne fruit in the production of this book. Surgeons in the past have neglected the œsophagus because the results of operative treatment in malignant disease were so poor. However, to-day, with improved technique, the modern methods of anæsthesia, the use of antibiotics, and blood transfusion, the surgeon can perform operations which would have proved fatal a decade ago.

This monograph, written by a surgeon with an extensive and mellowed clinical background, gives the reader an up-to-date picture of the surgery of the œsophagus. This includes the treatment of congenital atresia, strictures, pouches, and diverticula, and carcinoma of the œsophagus.

We congratulate the author on a first-class useful monograph.

A Survey of Cancer in London. Report of the Clinical Cancer Research Committee, by W. L. HARNETT, C.I.E., M.D., F.R.C.S., Lt.-Col. I.M.S. (Ret.), Medical Secretary to the Committee. With a Foreword by the Rt. Hon. LORD HORDER, G.C.V.O., M.D., F.R.C.P., and an Introduction by Sir HENEAGE OGILVIE, K.B.E., D.M., M.Ch., F.R.C.S. 7¼ × 9¾ in. Pp. 834 + vi, with 22 illustrations. 1952. London: British Empire Cancer Campaign. Cloth 50s.; Paper 45s.

IN the Foreword Lord Horder states that this report "deals mainly with the question of differential treatment and its results". This is obviously of vital importance and should serve as a guide for the best treatment available for a growth in a particular organ. As the result of research into the treatment, it is becoming more and more obvious that each case demands a special study by a trained eye and mind to decide which form of treatment holds out the greatest hope for success.

The importance of this cannot be over-estimated as it is *the first treatment* which is the most important. To be successful it must be carried out sufficiently early, and adequately by those whose past experience and training is such as to enable them to give the best. This will never be achieved if it should be solely at the discretion of a so-called 'cancer specialist'.

Sir Heneage Ogilvie in the Introduction to this report states: "The objects for which the British Empire Cancer Campaign has been established are stated in the Memorandum of Association in the following terms", namely, "to attack and defeat the disease of cancer in all its forms, to investigate its causes, distribution, symptoms, pathology and treatment, and to promote its

cure". To do this adequately implies teamwork of the highest order.

It is necessary, therefore, to have accurate information "about the methods of treatment available, and about the proportion of cases suffering from cancer of different types that is cured by each method, alone or in conjunction with other methods".

To obtain this in 1935 the Campaign appointed a Clinical Cancer Research Committee, who decided in 1936 to confine the inquiry to London and furthermore :—

1. "That an effort should be made in each hospital to co-ordinate all the cancer material in that hospital by a group of clinicians with one co-ordinating clinician.

2. "That there should be a paid Cancer Registrar in each hospital.

3. "That in each hospital there should be, in addition to the ordinary hospital notes, a separate case record for each cancer patient and that these case records should be available for use by this Committee.

4. "That it is advisable to establish a Cancer Follow-up Department in each hospital, the records of which should be maintained by the Cancer Registrar and should be available for the use of this Committee."

That the public is becoming cancer conscious is obvious by the fact that questions are now being asked in the House of Commons about 'cancer education' of the public. Many even go so far as to suggest that cancer should be a notifiable disease. This would undoubtedly produce 'cancer dread' quite apart from the fact that it is so difficult to diagnose as there is no definite symptomatology, or, as one author has stated : "In many instances the diagnosis of cancer can be made with certainty, in a much greater proportion of cases the opinion of a consultant reinforced by radiological and pathological examination is essential before a *possible* case of cancer becomes a *probable* one. Further, in many cases a surgical operation may be necessary before a *probable* case becomes a *proved* one."

It is for these reasons that "The Ministry has been advised against a cancer campaign addressed directly to the public", but they must be kept informed of the progress in combating the disease. It is for this purpose that The Imperial Cancer Research Fund, and the British Empire Cancer Campaign were inaugurated, and hence the extreme value of such a report as the present.

The education of the public must be achieved through *Hope* rather than by *Fear*.

Propaganda is not necessary ; facts can be supplied accompanied by hope and encouragement. It was proved by Mackenzie (1939) that a patient delays seeking advice owing to ignorance, fear, gullibility, false modesty, or concomitant disease.

The family doctor should be kept up to date with all the results of research in making as early a diagnosis as possible, and to do this he must have the necessary time to take a complete history and make a thorough clinical examination of each case. An adequate number of beds must be available, and facilities for consultation and investigation be easily obtainable, without long journeys and enforced absence from home in these difficult times. The family doctor is the principal witness whereby an adequate 'follow-up' system can be established.

Some centralization for treatment is necessary to secure an adequate first treatment. Apparatus is becoming more specialized and costly, hence from an economic point of view it must be centralized and when installed work at a capacity that prevents waste. Registration can be more efficiently controlled and kept up to date by a central organization.

It is quite impossible to review in any detail a report such as this. It demands and is worthy of a special study by all those who are brought into contact with malignant disease, and especially so if called upon to treat it. This really includes the whole of the medical profession. The treatment does not cease when no further curative or palliative procedures can be carried out ; for it is during the subsequent period, until the patient is relieved by death, which often calls for the most skill in the art of medicine.

Furthermore, much besides the giving of sedatives will be possible in the future to relieve pain.

The report begins with a short summary which details the various headings under which the survey was made, such as number of cases, anatomical site, and sex and age distribution, and relevant factors such as heredity, past history, occupation, and social class. Then symptoms are considered in priority of appearance and the time interval between the appearance of them and the time of going to the doctor. The advice given by the family doctor is contrasted with the physical signs of the lesion and general condition of the patient when admitted to hospital ; and finally the treatment given and the results of a five-year follow-up.

It has all been done with meticulous care and no exaggerations.

A separate chapter is devoted to each site in the body where a growth may occur.

The final chapter is headed E. & O.E., which means errors and omissions excepted. This is an example which might well be followed by others who compile statistics with sometimes such obvious satisfaction to themselves, but not to their readers. There is also an index of the authors who have been cited, and finally what is so very necessary in such a report – a very comprehensive index.

The clinical committee and the Campaign are to be congratulated on this report. It is a model of its kind ; it is long, but not too long for the comprehensive way the whole subject is covered.

The Medical Secretary, Col. W. L. Harnett, must be extremely proud of this culmination to all the work, time, and thought he has obviously given to its production ; and he must have inspired his assistant staff with enthusiasm sufficient to enable it to have been made.

Every hospital dealing with malignant disease should have the volume for reference, so that the staff may appreciate the importance of accurate registration and follow-up of these cases. It would give them an opportunity to play an important part in waging warfare against this disease, one in which no quarter is to be asked for or given, and no armistice can take place. The only result must be unconditional surrender and absolute victory.

Prostatectomy. A Method and its Management. By CHARLES WELLS, Professor of Surgery, The University of Liverpool. $6\frac{3}{4} \times 9\frac{7}{8}$ in. Pp. 103 + viii, with 72 illustrations. Edinburgh : E. & S. Livingstone Ltd. 24s.

IN this delightful monograph Professor Wells has recorded the methods used and results obtained at his clinic in 200 consecutive cases of prostatic obstruction. As is generally known, he is a staunch advocate of the Wilson Hey operation, and although he does not claim to speak for the originator he has produced results with which Mr. Wilson Hey may well be pleased.

The historical, pathological, and clinical aspects are dealt with in a manner unlikely to cause much dissension ; we consider that a distended bladder will accentuate the degree of prostatic enlargement as estimated on rectal examination rather than the reverse.

The most important special investigation used is intravenous pyelography, and it is carried out in every case, so that a grading by dilatation is possible. Should this really be done when the blood-urea is 200 ? In the case cited even a double dose of pyelectan failed to demonstrate secretion, and that is the usual experience.

The main controversy will centre around the advisability of 'immediate' prostatectomy in patients with chronic retention. There is no doubt that it can be

successfully performed in many cases, and the figures quoted here are ample evidence thereof. There are, however, many patients with chronic retention who are 'gravely ill' from causes other than the prostate, and for them the treatment advised is catheterization. It is surely in this group that drainage by suprapubic catheter will allow of medical treatment for the intercurrent illness, making prostatectomy possible at a later stage. The operation itself has not found universal favour, but is evidently satisfactory; it appears to have the disadvantages of needing a finger in the rectum and of taking two hours to perform. The experienced urologist is unlikely to change his well-tried methods in favour of it.

There may be critics of the method, but there can be none of the manner of its presentation. Professor Wells has done a great service to surgery in putting his views on record; if they represent one extreme of the swing of the pendulum they will at least help to combat the undue conservatism which makes a two-stage Freyer the routine procedure in some quarters.

Renal Diseases. By E. T. BELL, M.D., Professor of Pathology in the University of Minnesota. Second edition. 5¾ × 9 in. Pp. 448, with 123 illustrations and 4 colour plates. 1950. London: Henry Kimpton. 56s.

THE second edition of this valuable book has been brought up to date by the addition of new work. It is appropriate that an account which includes both medical and surgical diseases should be written by a pathologist, although perhaps inevitable that the medical aspect should receive more detailed attention than the surgical.

We agree with the author that the term 'lower nephron nephrosis' has added confusion instead of clarity, and should be abandoned. We do not agree with his criteria of prognosis in adenocarcinoma of the kidney; there is no mention of the histological grading of these tumours, which together with the presence or absence of involvement of the renal vein is of more importance than the size of the tumour in assessing prognosis.

This edition gives a very lucid and complete account of diseases of the kidney. The section on hypertension is of particular interest.

The 1951 Year Book of Urology (November, 1950-October, 1951). Edited by WILLIAM WALLACE SCOTT, M.D., Ph.D., Director, James Buchanan Brady Urological Institute, The Johns Hopkins Hospital; etc. 5 × 7⅞ in. Pp. 383, with 84 illustrations. 1952. Chicago: Year Book Publishers Inc. London: Interscience Publishers Ltd. 42s.

YEAR by year the value of this annual publication increases. It gives an admirable summary of much of the important literature on urology, and is a ready means of reference to the original article if necessary. This volume is enhanced by an account of the Editor's visit to Great Britain; he makes favourable comment on our methods of undergraduate teaching and on the increased interest in urology fostered by the formation of the British Association of Urological Surgeons.

BOOK NOTICES

[The Editorial Committee acknowledge with thanks the receipt of the following volumes. A selection will be made from these for review, precedence being given to new books and to those having the greatest interest to our readers.]

Surgical Care. A Handbook of Pre-operative and Post-operative Treatment. By RONALD W. RAVEN, O.B.E. (Mil.), F.R.C.S., Hunterian Professor, Royal College of Surgeons; Joint Lecturer in Surgery, Westminster Medical School; Surgeon to Westminster (Gordon) Hospital; Surgeon to the Royal Cancer Hospital. Second Edition. 5½ × 8½ in. Pp. 435 + xii, with 68 illustrations, a number as plates. 1952. London: Butterworth & Co. (Publishers) Ltd. 37s. 6d.

Diseases of the Chest. Edited by Sir GEOFFREY MARSHALL, K.C.V.O., C.B.E., M.D. (Lond.), F.R.C.P. (Lond.), Consulting Physician, Guy's Hospital; etc.; and KENNETH M. A. PERRY, M.A., M.D. (Cantab.), F.R.C.P. (Lond.), Assistant Physician, The London Hospital; etc. In two volumes. 6⅝ × 9¾ in. Vol. I. Pp. 456 + xii, with 19-p. index and 158 illustrations. Vol. II. Pp. 413 + viii, with 31-p. complete index and 192 illustrations. 1952. London: Butterworth & Co. (Publishers) Ltd. £7 7s.

Pictorial Handbook of Fracture Treatment. By EDWARD L. COMPERE, M.D., F.A.C.S., and SAM W. BANKS, M.D., F.A.C.S., Northwestern University Medical School. Third edition revised with the assistance of CLINTON L. COMPERE, M.D., F.A.C.S. Illustrated by HAROLD LAUFMAN, M.D., F.A.C.S. 5½ × 8 in. Pp. 424, with 223 illustrations. 1952. Chicago, Ill.: The Year Book Publishers Inc. 56s.

Human Actinomycosis. What the General Practitioner ought to know. By V. ZACHARY COPE, M.S., F.R.C.S., Consulting Surgeon, St. Mary's Hospital and the Bolingbroke Hospital. 4¾ × 7⅝ in. Pp. 80 + xii, with 32 illustrations. 1952. London: William Heinemann Medical Books Ltd. 12s. 6d.

Surgery for Students of Nursing. By JOHN CAIRNEY, D.Sc., M.D., F.R.A.C.S., Director-General of Health for the Dominion of New Zealand. With a Foreword by Miss M. I. LAMBIE, C.B.E., First Vice-President, International Council of Nurses. 5½ × 8½ in. Pp. 326 + x, with 120 illustrations. 1952. Christchurch, N.Z.: N. M. Peryer Ltd. N.Z. price, 40s.

Aids to Theatre Technique. By MARJORIE HOUGHTON, M.B.E., S.R.N., S.C.N., D.N. (Lond.), formerly Sister Tutor, University College Hospital, London. With a Foreword by D. R. DAVIES, F.R.C.S., Surgeon, University College Hospital, London. Second edition. 4 × 6⅜ in. Pp. 260 + xvi, with 125 illustrations. 1952. Baillière, Tindall & Cox. 6s.

Introduction to Dental Anatomy. By JAMES H. SCOTT, B.Sc., M.D., L.D.S., Lecturer in Anatomy for Dental Students, The Queen's University of Belfast, and NORMAN B. B. SYMONS, M.Sc., B.D.S., Lecturer in Dental Anatomy and Histology and Curator of the Dental Museum in the University of St. Andrew's at the Dental School, Dundee. 5½ × 8⅛ in. Pp. 292 + viii, with 172 illustrations. 1952. Edinburgh: E. & S. Livingstone Ltd. 35s.